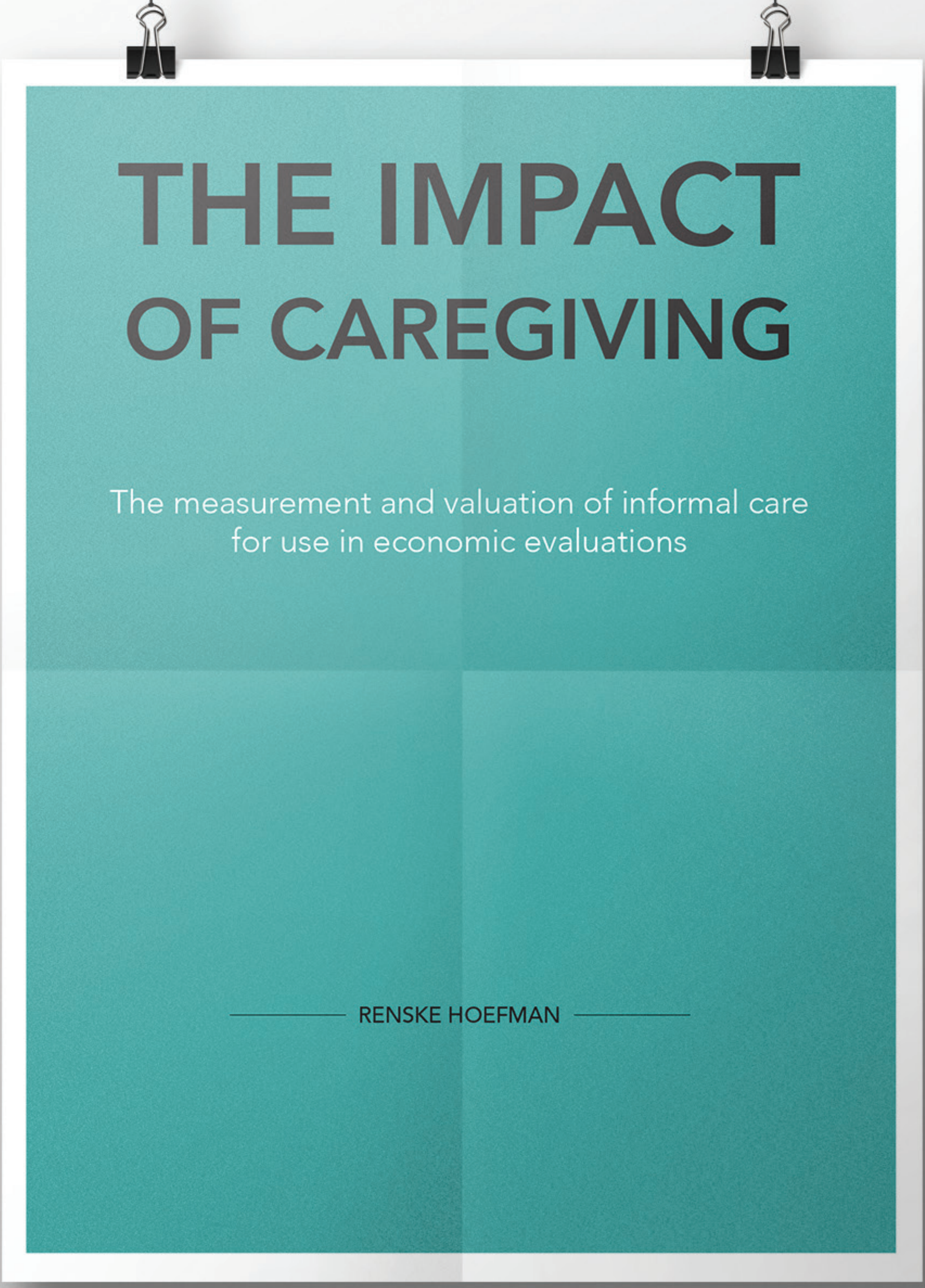

THE IMPACT OF CAREGIVING

The measurement and valuation of informal care
for use in economic evaluations

RENSKE HOEFMAN

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The measurement and valuation of informal care for use in economic evaluations

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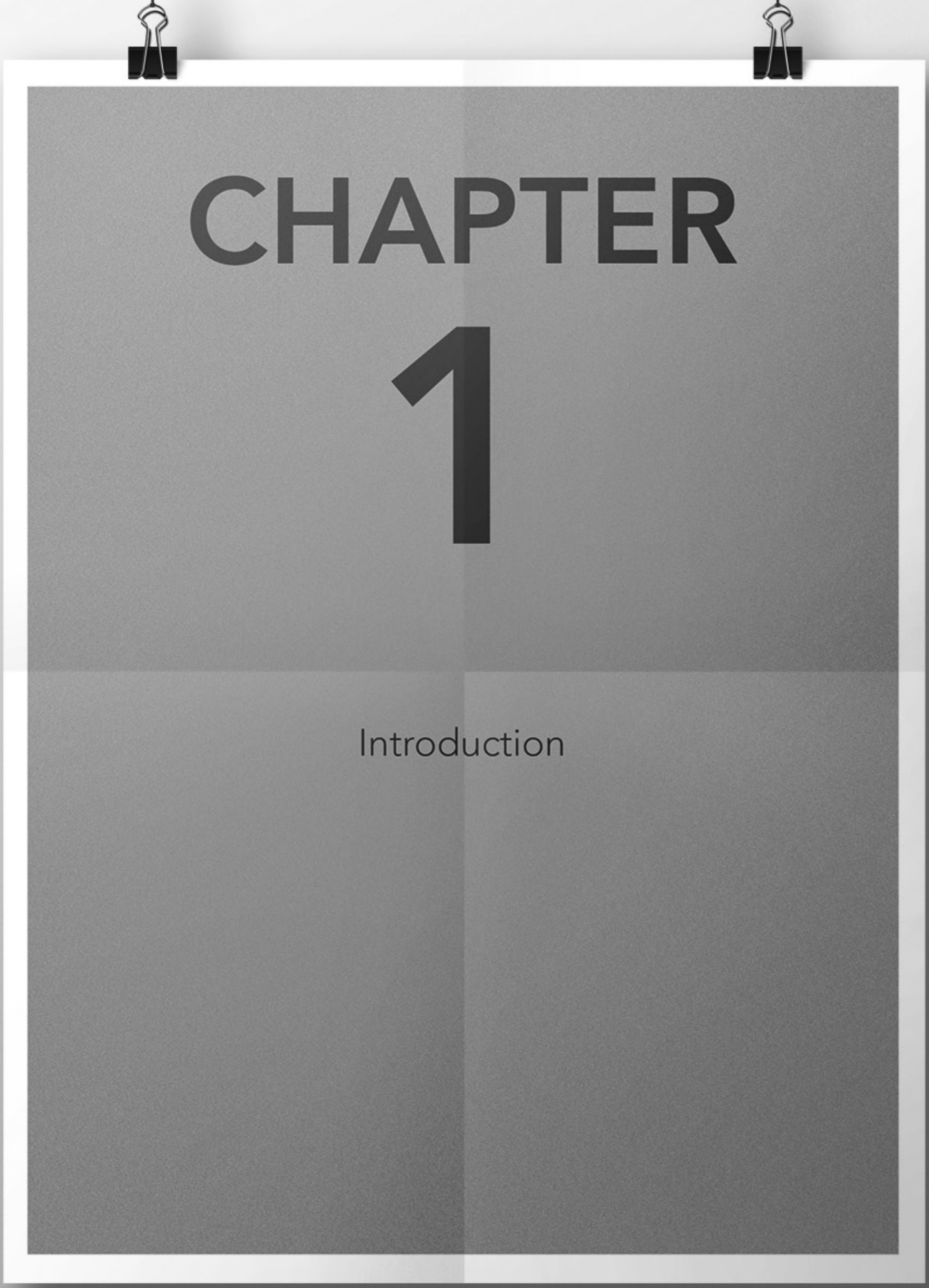


CHAPTER

1

Introduction

Families and friends often form 'the backbone' of care for patients with long term health needs. Roughly between 5 and 30 per cent [1-7] of the adult population in Western countries provide informal care. The exact figures differ between countries given cultural differences in family life and responsibility, differences in long-term care systems and different definitions of caregiving [3, 8, 9]. Central in most definitions of informal care is that (i) the need for care is caused by health problems, disability or infirmity due to old age, (ii) caregivers[1] and care recipients already had a social relationship prior to the start of caregiving, and (iii) that, although perhaps perceived as forced by the circumstances, care is provided on a voluntary basis and usually without financial compensation. The importance of informal care is emphasised by the fact that many caregivers lend care during several years. Moreover, while the majority of caregivers spend less than ten hours per week on informal care, a substantial proportion of caregivers provide many more hours of care to high care need patients [1, 3, 6, 10-13]. Caregivers usually perform a diversity of activities, such as assisting the patient with eating, dressing and personal hygiene, shopping, doing laundry and managing medication or finances [4, 6, 8, 13-16].

Notwithstanding the importance of informal care, it can be both mentally and physically burdensome for caregivers. For instance, in the context of patients with psychological disorders or in case of severe illnesses, providing care can be emotionally and mentally straining. Care can be physically demanding when caregivers repeatedly have to perform care tasks such as helping a patient in and out of bed or with bathing and clothing. Caregivers often have to combine care tasks with other responsibilities, such as looking after their children or performing paid work. Combining these different roles can lead to additional strain. It follows that caregivers often experience diverse problems, such as stress, depression or physical health problems. In the literature, the consequences of caregiving are usually described in terms of (i) objective burden, (ii) subjective burden, (iii) health, or (iv) well-being of caregivers. Figure 1.1 presents the conceptual model of the impact of caregiving on caregivers used in this thesis. This conceptual model is based on the stress process model of Pearlin et al [17] and the appraisal model of Lawton et al [18-20]. In this model, family and friends are confronted with a demand for care of the care recipient. The demand for care is determined by the health problems of the care recipient. For example, care recipients' mental and/or physical health problems, their independence in activities of daily living, and the need for continuous supervision are important determinants of the demand for care [11, 21-24]. Consequently, family and friends evaluate this demand for care and may decide to provide informal care. In Figure 1.1, the provision of informal care is described in terms of objective burden. Objective burden concerns the 'objectively' measurable characteristics, or inputs, of informal care. It consists of characteristics such as the care activities performed, the time invested in caregiving and the duration of caregiving [25-27]. The objective burden of caregiving may influence caregivers' lives in several ways. Important to note is that caregivers may respond differently to a similar level of objective burden of caregiving. As depicted in Figure 1.1, the objective burden of caregiving may influence caregivers' subjective burden, health and/or well-being. First, subjective burden concerns the strain from caregiving as experienced by caregivers [26]. It is well-established that caring can be straining, even though most caregivers consider it to be a natural consequence of their relationship with the care recipient [14, 28]. The level of subjective burden that caregivers experience depends on their evaluation of the positive and negative effects of caregiving and their ability to cope with these effects [25, 29]. Examples of positive effects of informal care are improving the relationship with the care recipient, the feeling of performing a meaningful task, acquiring (management) skills, increasing knowledge of health

---

[1] In this thesis, we refer to informal caregivers whenever we use the word 'caregiver'.

issues, experiencing personal growth and obtaining more appreciation for everyday life [14, 28, 30-33]. Negative effects of caregiving may be diverse as well and include factors such as disruptions of personal or social lives, marital problems, family conflict and social withdrawal due to caregiving [13, 21, 24, 33-37]. Furthermore, caregivers may face financial strain or even increased risk of poverty due to out-of-pockets expenses related to caregiving or limitations in labour participation [3, 6, 14, 38-41]. Secondly, providing informal care may have health consequences for caregivers [11, 13, 42, 43]. While some studies report certain positive health effects of caring, such as decreased depression or anxiety over time [44], most seem to indicate that caregivers may experience considerable health problems. For example, caregivers may experience psychological symptoms, such as stress, depressive disorders or anxiety. They may also experience physical health symptoms, leading to higher medication use and hospitalization, while elderly caregivers even seem to have higher mortality rates [13, 42, 45-52]. Thirdly, studies also report consequences of caregiving in terms of lower general well-being or overall quality of life [28]. Providing informal care may negatively impact diverse aspects of caregivers' overall quality of life, such as their physical, material, social and emotional well-being [21, 27, 28, 46, 53, 54].

The four outcomes of caregiving (*objective burden, subjective burden, health and well-being*) are influenced by characteristics of the care recipient, the caregiver and the care situation (see Figure 1.1) [55]. Examples of socio-demographic variables of caregivers associated with consequences of caregiving are the caregivers' age, gender, educational level, partner status, household composition, income level, and (un)paid work position [13, 21, 33, 34, 39]. Moreover, age and gender of care recipients and care situation characteristics such as the relationship between the caregiver and care recipient, whether the caregiver is the primary caregiver, the living situation of the care recipient, the receipt of formal care or support and the motivation to care are also important in this context [13, 22, 33, 34, 34, 39, 56, 57].

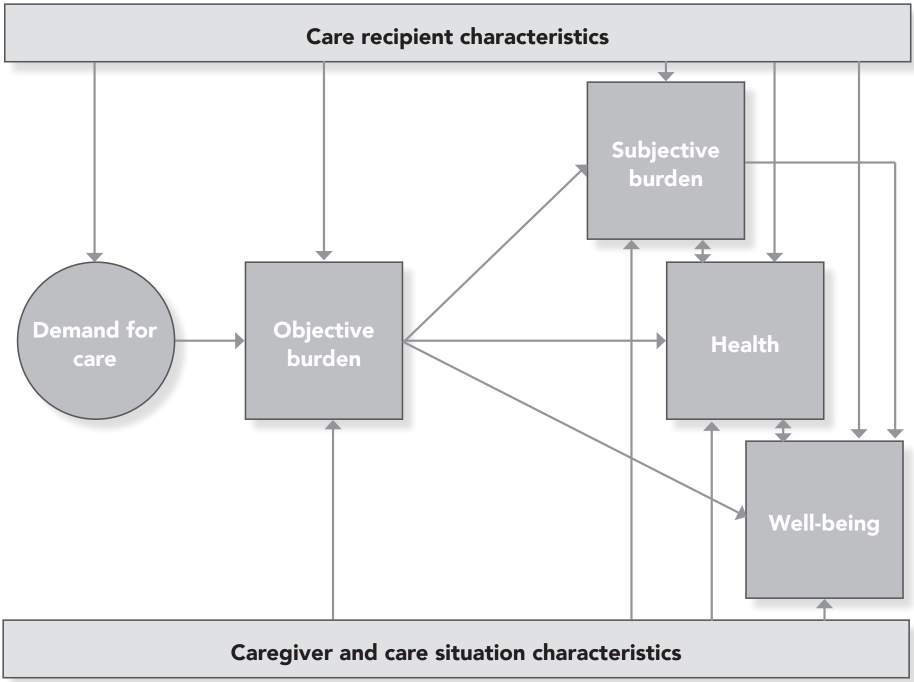


Figure 1.1 Conceptual model of the impact of caregiving on caregivers

1.1 Supporting long-term participation of caregivers

It is often questioned whether caregivers will be able to maintain caregiving to a sufficient extent in the future [29, 56, 58, 59]. Demographic and labour market developments in many Western countries, such as a scarcity of formal health care personnel and the rapid ageing of the population, suggest that the demand for informal care will increase in the coming years [3, 60]. Government policies also influence the demand for informal care. Policy makers often consider informal care to be an attractive alternative to formal care, because the associated costs do not fall on the formal health care budget. As a result, shifting care tasks from formal to informal care may reduce the pressure on formal care budgets. This occurs for instance when caregivers increasingly need to assist formal care personnel with their care tasks or have to take over these tasks [61-68]. At the same time, the number of persons available for informal care provision may be expected to decrease in the coming years, for instance due to increased labour participation of women and increasing geographical spread of families. To enable long-term participation of the available caregivers, it is crucial that they are effectively supported in their caregiving tasks. Nowadays, most Western countries provide support services for caregivers. Examples of these are respite care, care leave or flexible work arrangements to better balance caregiving, work and family life. Some Western countries also financially support caregivers. For instance, caregivers may receive money from caregivers' allowances or can be paid from the patients' cash benefits to manage care delivery. Whether caregiver support is effective largely remains uncertain for policy makers [2, 3]. This is problematic, since they are required to make choices about which interventions to finance from limited public budgets. More in general, choices in the provision of health care interventions typically lack attention for informal care. In health care, policy makers can base their funding decisions on several criteria. For example, the aim to distribute services fairly within a country is often mentioned [69]. Moreover, policy makers can question whether an intervention provides value for money or, in other words, whether it is cost-effective. Funding decisions on health care programmes are increasingly guided by cost-effectiveness considerations [70-72]. This especially applies to pharmaceuticals. However, it has been claimed that in health care sectors such as the long-term care sector [73], cost-effectiveness information should also be used in decision-making.

1.2 Informal care in economic evaluations of health care interventions

Cost-effectiveness information can be obtained through economic evaluations comparing the costs and effects of an intervention to its best alternative. Different types of economic evaluations exist, which can be distinguished by their unit for measuring the benefits of interventions [70, 74]. First, cost-benefit analysis (CBA) values the benefits of health care in monetary terms. Secondly, in cost-effectiveness analysis (CEA) gains of interventions are described in natural units, such as life years gained or reduction of disease activity. Thirdly, cost-utility analysis (CUA) uses 'utilities', a preference based measure combining length of life and health-related quality of life into quality-adjusted life years (QALYs). Lastly, cost-consequence/multi-criteria analysis (MCA) reports effects of interventions in terms of several relevant benefit measures to inform policy makers [70, 74].

When economic evaluations are used to inform policy making, it is important to incorporate all relevant costs and effects related to an intervention in the calculations. Which costs and effects are deemed relevant for cost-effectiveness considerations depends on the perspective of the economic evaluation [74-76].

Often, the societal perspective is recommended. The societal perspective implies that economic evaluations use a broad scope, indicating that all costs and effects that occur in society as a whole are relevant [29, 74, 75, 77, 78]. Hence, it is important that policy makers are not only informed about the costs of formal health care and the consequences for patients when making policy decisions in health care, but are also aware of the broader positive and negative consequences of interventions for society [37, 79-83]. For example, health interventions may not only affect patients, but may also influence persons in their social network [26, 29, 53, 84]. This can occur in different ways. A 'family effect' occurs when the health status of a patient directly affects the health or well-being of their family or friends (because they 'care about' the patient) [29, 50, 53, 58, 81, 85-88]. Moreover, interventions aimed at improving the health of patients may also have spillover effects on their caregivers, known as the 'caregiving effect', which occurs because they care for the patient [27, 29, 50, 53].

From a societal perspective, informal care resources entail real costs to society, even though these resources are not traded on a market, are not necessarily publicly financed and, hence, are less visible for policy makers [76, 79, 89, 90]. Costs of caregiving include (i) the opportunity costs of time caregivers spend on informal caregiving, i.e., the value of activities forgone like paid labour, and (ii) out-of-pocket expenses associated with caregiving, such as house adaptations or travel costs [74, 91] (see Figure 1.2). Next to all relevant informal care costs, it is important to include all effects related to informal care in economic evaluations in order to make a full assessment of welfare changes [74, 75, 77, 78]. Changes in (subjective) burden may result in changes in health or well-being of caregivers (see Figures 1.1 & 1.2).

Economic evaluations can also be conducted from a narrower perspective than the societal perspective, such as the health care perspective. This is currently recommended in several countries, for example the UK [74, 76]. The health care perspective typically restricts the inclusion of costs to those that fall under the health care budget (and sometimes other public budgets) and the effects to health effects. This implies that inclusion of informal care is only relevant information in specific circumstances. That is, if the care situation affects the health of caregivers (potentially leading to a demand for formal care), these effects can be included [25, 76, 82].

At present, informal care is often ignored in economic evaluations, even those claiming to adopt a societal perspective [29, 74, 75, 77, 78, 92]. Moreover, the few economic evaluation studies that do include informal care often only cover a part of the impact of caregiving [90, 91]. Excluding (specific consequences of) caregiving may bias cost-effectiveness information and therewith may lead to questionable policy recommendations [93]. For example, savings of health care programs resulting in early discharge of patients from hospital may be overestimated if the costs of the required extra informal care at home are not considered [94, 95].

Methodological challenges in quantifying the costs and effects of informal care may contribute to the neglect of informal care in economic evaluations [25, 91, 92]. Not only the measurement and valuation of informal care can be challenging, but also the inclusion of the outcomes in conventional health economic evaluations. In terms of measurement and valuation, it can for instance be difficult to validly measure actual time investment of caregivers or to value the full impact of informal care in monetary terms [91, 96, 97]. Instruments measuring and valuing the consequences of caregiving on caregivers for use in economic evaluations therefore ideally do not only (i) describe the consequences of an intervention in a feasible, reliable and valid way, but, in order to include it in conventional economic evaluations alongside patient effects, also (ii) value costs or benefits in a common currency, such as money in CBA or QALYs in CUA [70, 74]. If other outcome measures are used, such as subjective burden or well-being, it is difficult to combine outcomes in caregivers with conventional ways of expressing effects in patients, hampering inclusion in economic evaluations.

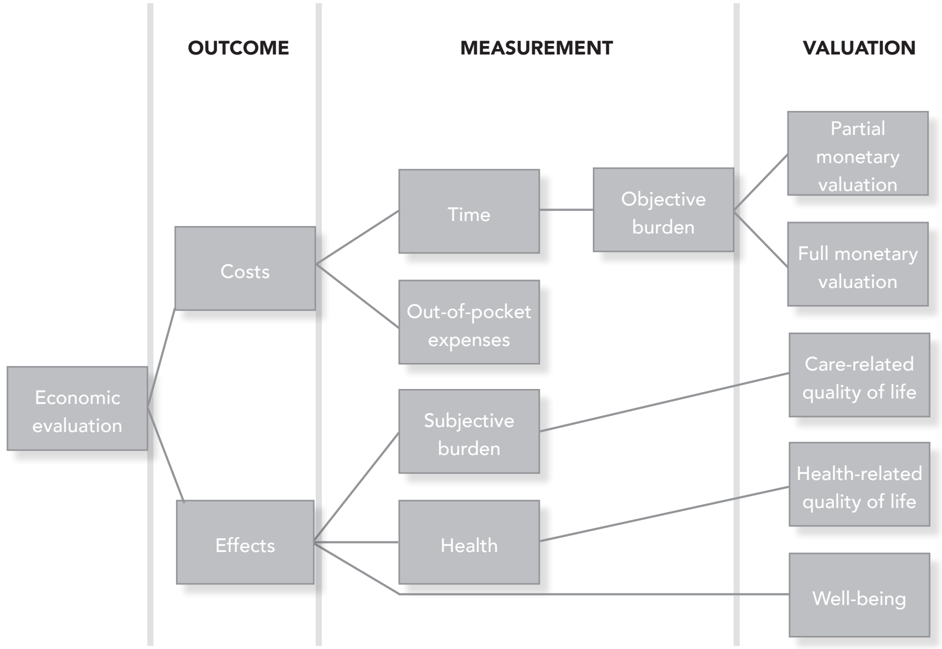


Figure 1.2 Costs and effects of informal care for inclusion in economic evaluations

1.2.1 Feasibility, reliability, and validity

Psychometric criteria, such as practicality, reliability and validity, can be used to assess whether instruments adhere to the first requirement of a feasible, reliable and valid instrument [70, 98]. First, practicality refers to whether the content and administration method is acceptable to respondents. One way of testing practicality is studying the feasibility of an instrument in terms of the proportion of surveys with completed responses [70, 98]. Secondly, reliability focuses on the reproducibility of the results of the instrument. For example, this psychometric criterion expects that dimensions of an instrument measuring the same underlying construct are correlated with each other (internal consistency). Moreover, test-retest reliability addresses the stability in responses on an instrument by the same respondents over time if the subject of measurement did not change [70, 98]. Finally, whether the instrument measures what it is intended to measure can be investigated through validity testing. Criterion validation compares the instrument to a gold standard. However, in social sciences such a gold standard is often non-existent. Alternatively, construct validity can be investigated. Different types of construct validation exist. For example, convergent validation examines whether an instrument correlates with other instruments measuring the same construct. Discriminative validation focuses on the ability of an instrument to differentiate between relevant groups of respondents.

Furthermore, clinical validation tests whether a scale is associated with variables important in the underlying theory of the instrument's construct [70, 98].

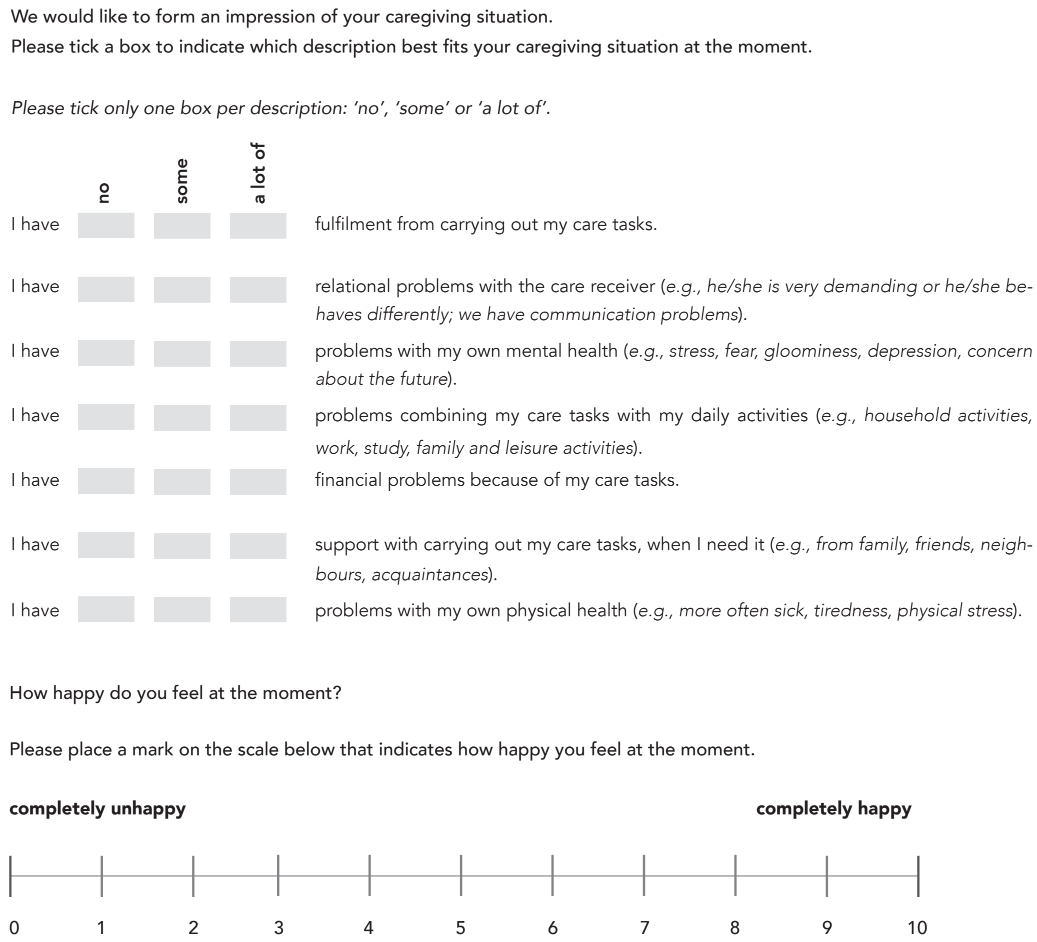
1.2.2 Valuation of costs and effects

Informal care can be valued in monetary terms and, subsequently, be included as a cost in economic evaluations. When valuing informal care as a cost, typically the time spent on caregiving is measured. Usually, this time is measured in terms of hours of caregiving per week with objective burden instruments. Next, the hours of caregiving are multiplied with a specific monetary value per hour [91]. This monetary value can be obtained through different monetary valuation methods, which provide either a partial or full monetary valuation of the impact of caregiving on caregivers [25, 26] (see Figure 1.2). Advantages of monetary valuation methods are that the required information can relatively easy be gathered and the results can be straightforwardly included at the cost side of economic evaluations. However, some of the monetary valuation methods only cover a part of the impact of caregiving, while the validity of some of these methods has been questioned [25, 29, 34, 91, 99-101]. Moreover, when using monetary valuation methods, it is doubtful whether policy makers in health care are sufficiently informed about the diverse impacts caregiving can have on caregivers. That is, the consequences of caregiving may become less visible in cost-effectiveness information. The costs associated with caregiving become part of the other cost components generally included in economic evaluations. Hence, no explicit attention is paid to the care situation or the type of problem experienced by caregivers [25, 26].

Rather than including informal care as a cost, it can also be incorporated at the effect side of an economic evaluation (see Figure 1.2). Different non-monetary (valuation) methods are available per type of effect: subjective burden, health or well-being (see Figure 1.2). Starting with the broadest outcome, well-being of caregivers can be seen as a reflection of individual welfare [102]. Well-being as an outcome measure in the context of caregiving has the advantage that it encompasses all domains of quality of life, such as health, social relationships and level of independence [26, 53]. Hence, well-being can include all possible effects caregiving may have on diverse aspects of caregivers' lives. However, the multi-dimensional nature of well-being inherently has disadvantages as well. For instance, effects related to life in general and not directly to caregiving, such as income or marital status [102], will also affect well-being scores. Therefore, it has been questioned in the literature whether this outcome might be too broad in the context of measuring consequences of informal caregiving for inclusion in economic evaluations [53, 103]. Secondly, health effects in caregivers can be valued with the previously mentioned health-related quality of life using the concept of QALYs [47], as usually applied in CUA in health care. However, the focus on a single domain of quality of life, i.e., health-related quality of life, might be insufficient to capture all consequences of caregiving. It is possible that effects from caregiving in other potential important domains of quality of life than health only, such as social relationships or financial stability [25, 26], will then be ignored. Hence, using health effects may lead to a partial valuation of the impact informal care. Thirdly, subjective burden as an outcome of caregiving provides a direct focus on caregiving by covering diverse problems caregivers might encounter [26, 104, 105]. The often used Zarit Burden Interview (ZBI; [106]) and Caregiver Strain Index (CSI; [107]) are examples of instruments focusing on diverse problem dimensions. Some subjective burden instruments, such as the Self-rated burden scale (SRB; [105]) or Carer Experience Scale (CES; [108]), also cover positive aspects of caregiving. An advantage of subjective burden instruments is that they are very informative regarding the experiences of caregivers. However, they generally fail to provide an overall valuation of the effect of caregiving in an economic sense. To overcome this, preference-based scores of ‘care-related quality of life’ for subjective burden instruments have been developed (see Figure 1.2), making them ‘utility measures’. These scores take differences in the importance of problems in the care situation into account. Such care-related quality of life scores enable comparisons of care situations as well as comparisons of the effects of different interventions. Moreover, care-related quality of life scores can be applied in economic evaluations targeted directly at patients, their caregivers, or both. To date, only three instruments provide preference-based scores of ‘care-related quality of life’; the Caregiver Quality of Life Instrument (CQLI; [109]), the Carer Experience Scale (CES; [108]), and the Care-related Quality of Life instrument (CarerQol; [110]). The calculation of utility scores for the CQLI was pioneering work. To date, however, the CQLI is of limited practical use in economic evaluation studies. This is mainly due to methodological difficulties, such as the complexity of the valuation method. For example, the applied time trade-off method task is difficult to understand for respondents in the context of informal care. Moreover, it is rather costly to apply the CQLI, because trained nurses are needed for data collection [25, 26, 91]. The CES and the CarerQol both seem more promising in that respect. Both are survey instruments, which can be self-completed by informal caregivers who can indicate how they score on positive and negative domains related to caregiving. Both instruments value the different ‘care states’ that are described with the instruments. Nevertheless, at the time of the start of this PhD thesis both the CES and the CarerQol could not claim to offer a feasible, reliable and valid description and valuation of the effects in caregivers. For instance, only two studies showed encouraging results on construct validity of the CES [111, 112]. Moreover, the CES uses preferences of caregivers of elderly people in the UK for its valuation [108, 111, 113]. At the start of this PhD thesis, for the CarerQol, some psychometric properties, in specific feasibility as well as construct validity, had successfully been tested, but only in a relatively small population of caregivers [110]. Moreover, the CarerQol provided a valuation of the impact of caregiving on caregivers in terms of general well-being, but not yet in terms of care-related quality of life.

1.3 Research objectives

This thesis focuses on methodological aspects of including informal care in economic evaluations, with a special emphasis on the CarerQol instrument (see Figure 1.3). There are still several questions to be answered before the CarerQol instrument can be applied in economic evaluations to record the full impact of caregiving on caregivers. First, this thesis investigates psychometric properties of the CarerQol instrument as a measure of the effect of caregiving on caregivers by studying its feasibility, test-retest reliability and construct validity in more diverse samples of caregivers than the first validation study of the CarerQol [110]. Secondly, this thesis will derive standardized tariffs for CarerQol states based on preferences of the general public, which will allow computing care-related quality of life scores for care situations described by the CarerQol instrument. Finally, this thesis discusses the strength and weaknesses of several methods to measure and value informal care, including the CarerQol instrument.



We would like to form an impression of your caregiving situation.
Please tick a box to indicate which description best fits your caregiving situation at the moment.

Please tick only one box per description: 'no', 'some' or 'a lot of'.

	no	some	a lot of	
I have	☐	☐	☐	fulfilment from carrying out my care tasks.
I have	☐	☐	☐	relational problems with the care receiver (*e.g., he/she is very demanding or he/she behaves differently; we have communication problems*).
I have	☐	☐	☐	problems with my own mental health (*e.g., stress, fear, gloominess, depression, concern about the future*).
I have	☐	☐	☐	problems combining my care tasks with my daily activities (*e.g., household activities, work, study, family and leisure activities*).
I have	☐	☐	☐	financial problems because of my care tasks.
I have	☐	☐	☐	support with carrying out my care tasks, when I need it (*e.g., from family, friends, neighbours, acquaintances*).
I have	☐	☐	☐	problems with my own physical health (*e.g., more often sick, tiredness, physical stress*).

How happy do you feel at the moment?

Please place a mark on the scale below that indicates how happy you feel at the moment.

completely unhappy **completely happy**

Figure 1.3 The CarerQol instrument

Summarizing, this thesis deals with three research questions:

1. What is the feasibility, test-retest reliability, and construct validity of the CarerQol instrument as a measure of the effect of caregiving on caregivers in different caregiving contexts?
2. What are the preferences of the general public in the Netherlands for caregiving situations described by the CarerQol instrument?
3. How can informal care be included in economic evaluations of interventions in health care?

The outline of this thesis is as following:

Research question 1 is addressed in samples from different caregiving contexts: (i) caregivers from member registries of support centres in the Netherlands (chapter 2), (ii) caregivers of patients of a long-term care facility in the Netherlands (chapter 3), (iii) caregivers recruited through an online panel in the Netherlands (chapter 4), (iv) parents of children with an Autism Spectrum Disorder in the United States (chapter 5), and (v) caregivers of palliative care patients in Australia (chapter 6). In these chapters, feasibility (chapters 2, 3, 5, 6), construct validity (chapters 2-6), and test-retest reliability (chapter 3) of the CarerQol are studied.

Chapter 7 investigates what the relative importance is of different dimensions of caregiving described by the CarerQol instrument for the general public and uses this information to determine a tariff set to calculate care-related quality of life scores for care situations described by the CarerQol instrument (research question 2).

Chapter 8 discusses different methods to measure and value informal care (research question 3).

Chapter 9 reports the main findings per research question and discusses strengths and weaknesses of the research conducted in this thesis. Chapter 9 ends with research and policy recommendations.

CHAPTER

2

A new test of the construct validity of the CarerQol instrument: measuring the impact of informal caregiving

Based on Hoefman, R. J., van Exel, N. J. A., de Jong, S. L., Redekop, W. K., & Brouwer, W. B. (2011). A new test of the construct validity of the CarerQol instrument: measuring the impact of informal caregiving. Quality of Life Research, 20(6), 875-887.

Abstract

Purpose

Most economic evaluations of health care programmes do not consider the effects of informal care, while this could lead to suboptimal policy decisions. This study investigates the construct validity of the CarerQol instrument, which measures and values caregiver effects, in a new population of informal caregivers.

Methods

A questionnaire was distributed by mail (n =1,100, net response rate =21%) to regional informal care support centres throughout the Netherlands. Two types of construct validity, i.e., convergent and clinical validity, have been analysed. Convergent validity was assessed with Spearman's correlation coefficients and multivariate correlation between the burden dimensions (CarerQol-7D) and the valuation component (CarerQol-VAS) of the CarerQol. Additionally, convergent validity was analysed with Spearman's correlation coefficients between the CarerQol and other measures of subjective caregiver burden (SRB, PU). Clinical validity was evaluated with multivariate correlation between CarerQol-VAS and CarerQol-7D, characteristics of caregivers, care recipients and care situation among the whole sample of caregivers and subgroups.

Results

The positive (negative) dimensions of CarerQol-7D were positively (negatively) related to CarerQol-VAS, and almost all had moderate strength of convergent validity. CarerQol-VAS was positively associated with PU and negatively with SRB. The CarerQol-VAS reflects differences in important background characteristics of informal care, type of relationship, age of the care recipient and duration of caregiving were associated with higher CarerQol-VAS scores. These results confirmed earlier tests of the construct validity of the CarerQol. Furthermore, the dimensions of CarerQol-7D significantly explained differences in CarerQol-VAS scores among subgroups of caregivers.

Conclusion

Notwithstanding the limitations of this study, such as the low response rate, this study shows that the CarerQol provides a valid means to measure caregiver effects for use in economic evaluations. Future research should derive a valuation set for the CarerQol and further address the instrument's content validity, sensitivity and reliability.

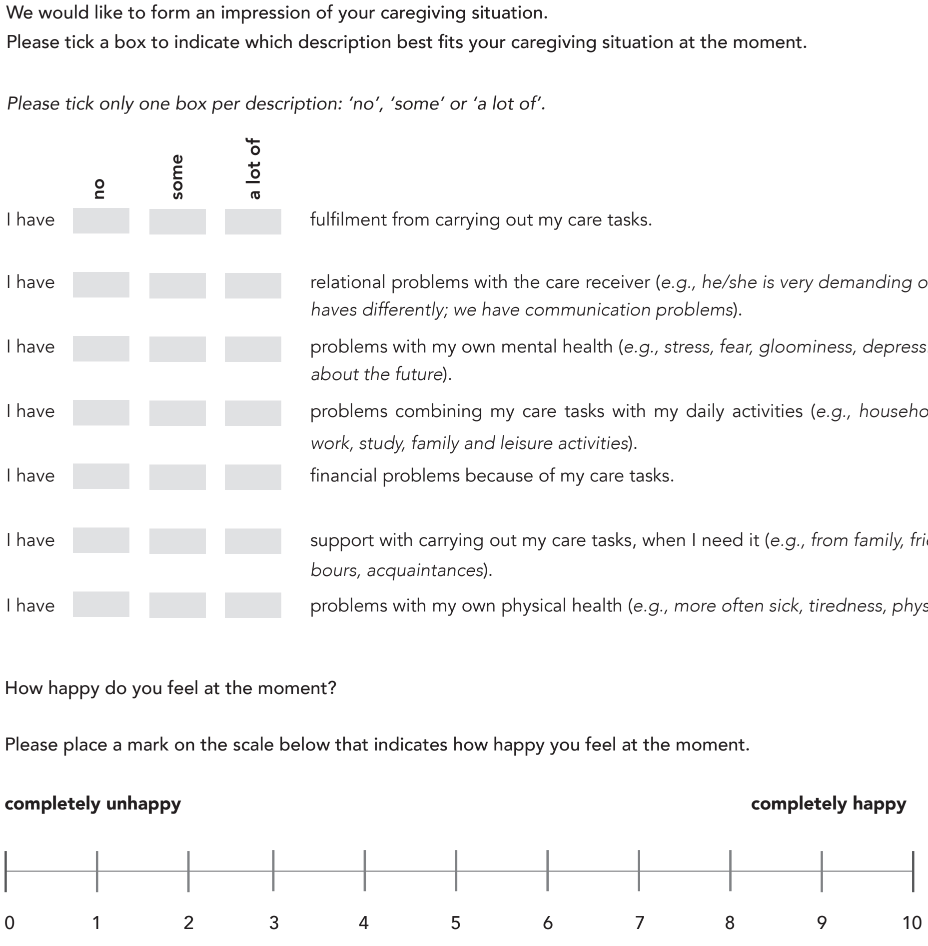
2.1 Introduction

Ill or disabled persons often largely rely on care provided by family or friends, typically non-paid. In the Netherlands, for instance, approximately 23 percent of the total population provides informal care [1]. Informal caregivers often do so over a long period of time and they spend a substantial amount of time per week on performing diverse activities which may be fairly demanding and unpleasant to perform. Consequently, informal care can be straining and has a profound impact on caregivers' lives. For example, informal care may result in problems to perform other activities, such as paid work or leisure activities. It can also lead to social isolation or financial strain, deterioration of a caregiver's physical and mental health and even increased mortality risk [14, 42]. Nevertheless, many caregivers consider the provision of informal care to their loved ones as a natural part of life, or as a simple obligation within a family relationship [14]. Moreover, informal care may have a positive impact on the well-being of caregivers. Many caregivers prefer to provide care to their ill or disabled loved ones themselves, rather than handing over care to someone else [28]. In addition, caregiving may enhance the quality of the relationship between caregiver and care recipient, increase the appreciation of everyday life, and stimulate the development of new skills [14], such as management skills to control the care situation.

The attention for informal care in economic evaluations of health care interventions seems to increase. Including informal care in such evaluations is particularly important if an economic evaluation claims to adopt the commonly advocated societal perspective, which entails the inclusion of all relevant costs and effects of an intervention, regardless of where these fall in society [74, 75, 77, 78]. Obviously, this includes the full impact of informal care. Health care interventions may not only affect patients, but may simultaneously influence their caregivers, either positively or negatively. For example, early discharge of patients from a hospital may save formal health care costs, but increase caregiver burden and, therefore, costs [95]. This increased burden moreover may in turn lead to more pressure on the health care budget when the health of the caregivers themselves deteriorates due to caring. In general, the exclusion of informal care in economic evaluations of health care interventions in which caregivers play a substantial role may thus lead to suboptimal policy recommendations. Even from a narrower health care perspective, as adopted in some jurisdictions (e.g., [114]), it can be argued that at least some elements of informal care are relevant, such as the health effects in caregivers [115]

To date, economic evaluations have usually failed to consider informal care [92]. When it is included the methods used often vary strongly between studies [91]. Typically, the opportunity costs or shadow price methods are used, which express the value of informal care in monetary terms by multiplying the number of caregiving hours with some value per hour [25, 99]. The resulting cost estimate can easily be included in an economic evaluation, i.e., on the cost-side of a cost-effectiveness ratio. A downside, however, is that such valuations do not necessarily reflect the preferences of caregivers [99], for example by not distinguishing between the first or the fiftieth hour of caregiving. Consequently, other monetary valuation methods have been proposed and applied, such as the contingent valuation method [116, 117], conjoint analysis (e.g., [118]) and the well-being method [119]. These methods supposedly are more sensitive to caregiver preferences, without losing the straightforward manner to include the results in common cost-effectiveness studies. In addition, non-monetary methods, traditionally mostly subjective burden measures, have been proposed to capture the impact of informal care (e.g., [120, 121]). Subjective burden measures indicate the burden experienced by caregivers.

However, while informative, such measures cannot be seen as valuation instruments in an economic sense; they are mainly descriptive. It has also been argued that, perhaps in addition to monetary valuation of time, the effects of informal care on health may be directly included in economic evaluations in terms of quality-adjusted life-years (QALYs) using validated questionnaires such as the EuroQol-instrument [122]. Changes in health-related quality of life of caregivers could then be combined with changes in patient health and included in the 'denominator' of a cost-effectiveness ratio [26]. Still, such an approach only comprises a partial valuation of informal care, i.e., it only captures the health effects of informal care. Hence, there is still the need for instruments that combine the information density of subjective burden measures with a comprehensive valuation method [108, 110]. This need gave rise to the development of the CarerQol instrument, acronym for care-related quality of life [110]. The CarerQol, which will be further discussed in the Methods section, comprises two parts (see Figure 2.1): a description of the care situation on seven burden dimensions (CarerQol-7D) and a valuation component in terms of general quality of life using a Visual Analogue Scale (CarerQol-VAS).



We would like to form an impression of your caregiving situation.
Please tick a box to indicate which description best fits your caregiving situation at the moment.

Please tick only one box per description: 'no', 'some' or 'a lot of'.

	no	**some**	**a lot of**	
I have	☐	☐	☐	fulfilment from carrying out my care tasks.
I have	☐	☐	☐	relational problems with the care receiver (*e.g., he/she is very demanding or he/she behaves differently; we have communication problems*).
I have	☐	☐	☐	problems with my own mental health (*e.g., stress, fear, gloominess, depression, concern about the future*).
I have	☐	☐	☐	problems combining my care tasks with my daily activities (*e.g., household activities, work, study, family and leisure activities*).
I have	☐	☐	☐	financial problems because of my care tasks.
I have	☐	☐	☐	support with carrying out my care tasks, when I need it (*e.g., from family, friends, neighbours, acquaintances*).
I have	☐	☐	☐	problems with my own physical health (*e.g., more often sick, tiredness, physical stress*).

How happy do you feel at the moment?

Please place a mark on the scale below that indicates how happy you feel at the moment.

completely unhappy **completely happy**

Figure 2.1. The CarerQol instrument

2.1.1 First test of the CarerQol instrument

Some psychometric properties of the CarerQol, in specific feasibility as well as construct validity, have been previously tested in a heterogeneous population of caregivers identified via regional informal care support centres in the Netherlands (n=175, [110]). Other psychometric properties of the CarerQol, such as reliability and sensitivity to changes, have not yet been established.

The first results of the psychometric properties of the CarerQol showed that it is a clear, easy to use and comprehensible instrument, of which the feasibility and construct validity, including the convergent and clinical validity, were good [110]. Increased burden measured in terms of the dimensions of the CarerQol-7D was associated with increased burden measured with other burden instruments, such as the Caregiver Strain Index (CSI, [107]) and the Self-rated Burden scale (SRB, [105]). Furthermore, CarerQol-VAS scores correlated well with the CarerQol-7D. More problems on CarerQol-7D dimensions led to lower CarerQol-VAS scores, while all effects were statistically significant except for one dimension ('support'; see Methods section for specific CarerQol-7D dimensions). Regarding the clinical validity of the instrument, the instrument discriminated well between groups [110]. Specifically, different characteristics of the caregiver, care recipient and care situation were related with CarerQol-VAS in the expected direction, e.g., caregivers having a partner, those in good health, those caring for their parent, and caregivers who care for persons who do not use professional day care had higher CarerQol-VAS scores.

2.1.2 Objectives and hypotheses

The central aim of this chapter is to further investigate the construct validity of the CarerQol using a new, but quite similar, population of caregivers. To achieve this aim, comparable methods to test convergent and clinical validity as Brouwer et al. (2006) were used. Convergent validity of the CarerQol is measured as its correlation with other instruments measuring the construct of subjective burden of informal care. Clinical validity concerns the extent to which differences in background characteristics, such as caregiver variables, are reflected in CarerQol-VAS scores. We further study the clinical validity of the CarerQol by performing subgroup analyses, highlighting the differences in CarerQol-VAS scores between groups of caregivers. Given the similarities in study samples, it was expected that the results on convergent and clinical validity would resemble those in Brouwer et al. (2006), i.e., that CarerQol-VAS scores are negatively related to increased burden measured with the CarerQol-7D and other subjective burden instruments. Likewise, a positive association between CarerQol-7D and these other instruments is expected. Concerning the clinical validity, it was expected that CarerQol-VAS would reflect differences in important background characteristics of informal care, such as the relationship between the caregiver and care recipient. Moreover, certain variables may have a different relationship with the CarerQol-VAS score in different subgroups of caregivers.

2.2 Methods

2.2.1 Population

Data were collected with written questionnaires distributed by mail in a heterogeneous population of informal caregivers contacted through regional support centres for informal caregivers throughout the Netherlands (n=1100). A regional support centre provides support to caregivers, e.g., by organizing self-help groups. The response rate was 25% (n=275), which is comparable to similar studies (e.g., [119]). In total, 230 cases were analysed; two cases were not suitable for further analysis, 43 were excluded due to missing items on the CarerQol or other important variables (net response rate=21%). More information on the dataset can be found in [100].

2.2.2. Measures

The CarerQol instrument was included in the written questionnaire (see Figure 2.1). The development of the CarerQol instrument was based on the approach used for the EuroQol-instrument [122]. The CarerQol-7D consists of seven dimensions describing the burden experienced by caregivers. These dimensions were selected based on a literature review of validated burden instruments and the completeness and clarity of this list was evaluated and confirmed in a small pilot study with a convenience sample of informal caregivers [110]. The CarerQol-7D includes (+/- indicating positive/negative dimension): fulfilment of caregiving (+), relational problems (-), mental health problems (-), problems with combining daily activities (-), financial problems (-), social support (+) and physical health problems (-) [110]. Respondents can indicate to what extent they experience problems in these dimensions in their care situation, range in possible answers: no, some, a lot. In total, 2187 (= 3^7) different care situations can be distinguished. The valuation component of the instrument (CarerQol-VAS) consists of a Visual Analogue Scale (VAS) measuring general quality of life in terms of happiness (see Figure 2.1); a broad measure of well-being or utility increasingly used in the field of (health) economics [123, 124]. Happiness scores are seen as 'the degree to which an individual judges the overall quality of his life-as-a-whole favourably' [125]. The CarerQol-VAS consists of a horizontal VAS indicating current happiness ranging from 'completely unhappy' (0) to 'completely happy' (10), with numeric anchors equally spaced between these two extreme levels. Previous research in the field of informal care showed that such a VAS is feasible and informative [28, 34] with results comparable to a five point verbal description of happiness [119]. This broad outcome measure makes the instrument sensitive to the variety of consequences informal caregivers may experience, e.g., health or financial problems. A disadvantage is that other not care-related aspects of life may also influence the happiness of a caregiver, e.g., income level and type of work.
The outcomes of the CarerQol can be included at the 'denominator side' in a multi-criteria or cost-consequence analysis. One can also view the instrument as a useful tool to perform a cost-utility analysis, if an intervention targeted specifically at caregivers is to be evaluated. More detailed information on the development of the instrument can be found in Brouwer et al. (2006).

In addition, the questionnaire included questions on background characteristics of caregivers, care recipients, and the care situation, including objective and subjective burden and process utility. Objective burden consisted of the duration of care in years and months, the intensity of care in days per week and hours per day, the type of care activities distinguishing activities of daily living (ADL), personal care, instrumental activities of daily living (IADL), the care recipients' need for constant surveillance, the use of professional home care, and the need and use of respite care. Respite care is a generic term for different informal care support interventions, e.g., day-care programs or support groups. Subjective burden was measured with a horizontal VAS (Self-Rated Burden scale; SRB) ranging from 'not straining at all' (0) to 'much too straining' (10) [105]. Process utility (PU) is the utility derived from the process of caregiving [28]. PU is computed by the difference in happiness between the current situation and a hypothetical situation in which all caregiving tasks would be taken over by a person selected by the care recipient and caregiver, in the home of the care recipient, free of charge.

2.3 Statistical analyses

2.3.1 Convergent validity

To test the convergent validity, the associations between CarerQol-7D and CarerQol-VAS were analysed using Spearman's correlation coefficients (Table 2.2). Further, to assess the relation between the two parts of the CarerQol instrument, CarerQol-VAS scores were predicted on the basis of the CarerQol-7D dimensions with the use of multiple linear regression analysis (Table 2.3). As discussed in the introduction, happiness is a broad outcome measure and therefore CarerQol-VAS may be influenced by variables not necessarily related to caring. To investigate this, the CarerQol-7D was related to a less broad outcome measure, i.e., subjective burden (SRB), also with the use of multiple linear regression analysis (Table 2.3). The CarerQol-7D dimensions were analysed as continuous variables in both models as additional analyses (not shown) showed that treating CarerQol-7D as a continuous or categorical variable produced similar results. In addition, the convergent validity was tested by the association between CarerQol-VAS and the two other measures of caregiver burden, SRB and PU, with Spearman's correlation coefficients (Table 2.2). Moreover, the same associations were tested for the CarerQol-7D (Table 2.2).

Different guidelines exist to assess the strength of correlation coefficients e.g., [126, 127]. In this study, the strength of the Spearman's correlation coefficients is indicated by the guideline of Hopkins [128]: <0.1 trivial; 0.1-0.3 small; 0.3-0.5 moderate; 0.5-0.7 high; 0.7-0.9 very high; >0.9 nearly perfect, which largely agrees with the other classifications. Correlation of small to moderate strength would be seen as a sign of validity, because happiness is a broad outcome measure and the CarerQol-VAS score may be influenced by more than the effects of caregiving alone, for instance by the 'mere' fact that a loved one is ill, i.e., 'family effect' [26, 53], and by other effects of life in general.

2.3.2 Clinical validity

Descriptive analyses of all variables were performed, in means/percentages. The bivariate relation between CarerQol-VAS and characteristics of caregivers, care recipients and care situation was assessed with One-way ANOVA tests (Table 2.1). Furthermore, the clinical validity of the CarerQol was tested in a combined dataset of the respondents of this study and of the respondents of the first test [110]. Using multiple linear regression analysis the CarerQol-VAS scores were first only related to the CarerQol-7D (as in Table 2.3) and subsequently, besides to the CarerQol-7D scores to additional variables describing caregiver, care recipient and care situation characteristics (Table 2.4). The same multiple linear model was then applied in subgroups of caregivers to further test clinical validity (Table 2.4). These subgroups consisted of caregivers with low or high SRB and caregivers with low or high PU (low SRB/PU: 33rd percentile, high SRB/PU: 66th percentile and higher). When using dummies, the reference categories of these variables were those with the highest CarerQol-VAS score. The CarerQol-7D dimensions were treated all as continuous in the regression models.

Statistical significance was based on an alpha error of 0.05, except in tables 3 and 4 where an alpha error of 0.10 was used due to a small number of respondents when analysing cases within categories of variables. Statistical analyses were performed using Stata® version 10.0 (StataCorp LP).

Table 2.1 Characteristics of the caregiver, care recipient and care situation (mean (SD) or percentages) and bivariate correlation with CarerQol-VAS, n=230

		% or mean (SD)	mean CarerQol-VAS	p-value [b]
Caregiver				
Age	<59 years	58.74 (12.74)	6.25	0.81
	≥59 years		6.31	
Gender	female	74.3%	6.15	0.08
	male	25.7%	6.65	
Educational level	primary/no	13.1%	6.22	0.88
	lower/middle vocational	61.6%	6.33	
	higher vocational/university	25.3%	6.19	
Paid work	full-time	7.8%	5.86	0.28
	part-time	20.0%	6.62	
	no	72.2%	6.23	
Relationship with care recipient	partner	44.9%	6.14	0.02
	parent(-in-law)	31.6%	6.29	
	child	10.2%	5.57	
	other	13.3%	7.08	
Subjective health	good	54.4%	6.77	0.00
	reasonable or bad [a]	45.6%	5.71	
Self-rated burden	<5	5.21 (2.70)	7.24	0.00
	≥5		5.76	
Process utility		0.30 (3.02)		
	positive	37.9%	7.23	0.00
	neutral	19.6%	6.65	
	negative	42.5%	5.25	
Care recipient				
Age	<68 years	68.04 (20.81)	6.19	0.59
	≥68 years		6.33	
Gender	female	52.0%	6.58	0.01
	male	48.0%	5.91	
Health (rated by caregiver)	good	11.3%	6.58	0.00
	reasonable	65.1%	6.56	
	bad	23.6%	5.36	
Continuous surveillance	yes	36.7%	5.87	0.02
	no	63.3%	6.49	
Care situation				
Total years care	<9 years	9.15 (9.37)	6.33	0.49
	≥9 years		6.14	
Days p/wk	<6 days	5.67 (2.17)	6.34	0.56
	≥6 days		6.17	
Hours p/wk	<42 hours	41.59 (44.75)	6.41	0.01
	≥42 hours		5.70	
Care activities (hours p/wk):				
adl-activites	<26 hours	25.89 (37.01)	6.42	0.00
	≥26 hours		5.49	
personal care	<16 hours	15.50 (29.89)	6.32	0.04
	≥16 hours		5.67	
iadl-activities	<12 hours	11.78 (24.19)	6.43	0.01
	≥12 hours		5.65	
Care recipient shares household	yes	55.0%	6.09	0.10
	no	45.0%	6.50	
Professional home care	yes	67.2%	6.24	0.68
	no	23.8%	6.35	
Day care	yes	20.3%	6.20	0.62
	no	79.7%	6.36	
Use respite care	yes	33.5%	5.82	0.01
	no	66.5%	6.52	
Need respite care	yes	51.3%	5.83	0.00
	no	48.7%	6.76	

[a] Reasonable health: 43.0% of respondents, mean CarerQol-VAS score: 5.75. Bad health: 2.6% of respondents, mean CarerQol-VAS score: 5.0

[b] One-way ANOVA test

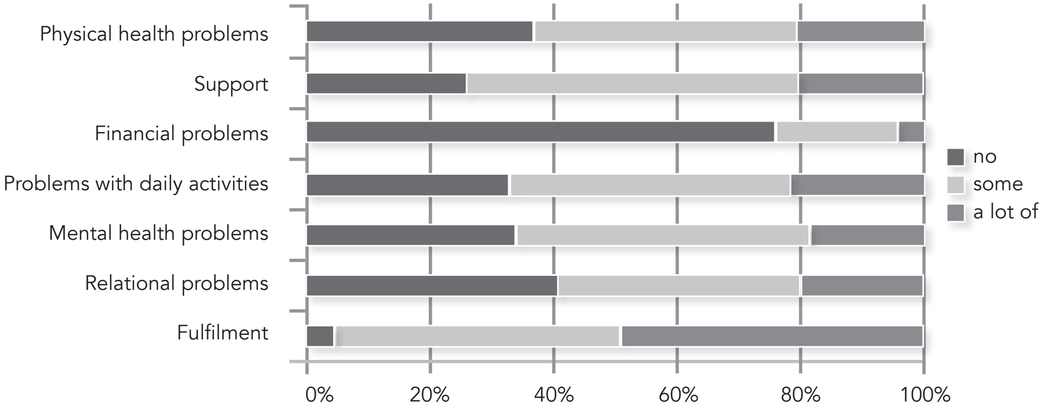
2.4 Results

Table 2.1 presents the characteristics of the study sample (n=230). Caregivers were 59 years, on average, and most of them were women. The majority of caregivers had a middle or higher educational level and approximately 28% was employed. Most caregivers provided care to a partner or a parent (in-law), and more than half shared a household with the care recipient, then often being their partner or child, not presented in table. Although the majority of caregivers was in good health, approximately 45% rated their health as reasonable or bad. On average, caregivers scored 5.2 on SRB. Mean PU was positive, meaning caregivers would become less happy when handing over all caregiving duties to someone else. Nonetheless, nearly 43% of all caregivers indicated that their well-being would increase in that case. Care recipients were predominantly women and, on average, 9 years older than the caregiver. Most had a poor health status and one out of three required permanent surveillance.

On average, caregivers had provided informal care for almost nine years. Currently, they provided care approximately 42 hours a week. Most time was spent on assisting the care recipient with personal care, e.g., eating and dressing, followed by household activities and support with practical issues, e.g., visiting friends and administrative tasks. Two out of three care recipients received professional home care and 20% used day care facilities. Respite care was used by more than one third of the caregivers and desired by approximately half of the other caregivers.

2.4.1 CarerQol-7D

Almost all caregivers experienced fulfilment from caring and the majority received support with their care tasks (Figure 2.2). Many caregivers had at least some relational problems with the care recipient or problems with combining caregiving with other activities. Almost two thirds reported physical and/or mental health problems. Although the majority did not have financial problems, one in four caregivers indicated having at least some financial problems due to caregiving.



Note: 8% of the respondents had one or more missing values on the CarerQol instrument

Figure 2.2 CarerQol-7D dimensions in percentages, n=230

2.4.2 CarerQol-VAS score

The mean CarerQol-VAS score was 6.3. Further, considering the bivariate relations (Table 2.1), it seems that happiness was highest among those caring for distant family or friends and lowest among those caring for their child. The scores were also higher when the health of the caregiver was good, when they felt less burdened and when their process utility was positive. Caregivers providing care to female care recipients, in relatively good health, and who do not need permanent surveillance, had significantly higher CarerQol-VAS scores, as did those who provided care less than 42 hours per week. Caregivers who did not need or use respite care reported higher happiness scores than those using or wanting to use respite care.

2.4.3 Convergent validity

The Spearman's correlation coefficients of CarerQol-VAS, SRB, PU and CarerQol-7D are shown in Table 2.2. CarerQol-VAS was negatively associated with caregiver burden (SRB score) and the CarerQol-7D dimensions relational problems, mental health problems, problems with daily activities, financial problems and physical health problems (range absolute values correlation coefficients 0.19-0.56). A positive association was observed with PU and the CarerQol-7D dimensions fulfilment and receiving support, although the latter was not statistically significant (range absolute values correlation coefficients 0.04-0.58). SRB and PU were associated with the CarerQol-7D dimensions in the expected way (range absolute values correlation coefficients 0.03-0.55).

Table 2.2 Correlation CarerQol-VAS and CarerQol-7D with Self-Rated Burden (SRB) and Process Utility (PU), (Spearman's rho; 2-tailed), n=230

	CarerQol-VAS	p-value	SRB	p-value	PU	p-value
SRB	-0.45	0.00				
PU	0.58	0.00	-0.44	0.00		
CarerQol-7D						
Fulfilment	0.23	0.00	-0.27	0.00	0.37	0.00
Relational problems	-0.34	0.00	0.36	0.00	-0.37	0.00
Mental health problems	-0.56	0.00	0.43	0.00	-0.35	0.00
Problems with daily activities	-0.44	0.00	0.55	0.00	-0.45	0.00
Financial problems	-0.19	0.00	0.23	0.00	-0.19	0.00
Support	0.04	0.57	-0.03	0.64	0.15	0.02
Physical health problems	-0.44	0.00	0.43	0.00	-0.39	0.00

2.4.4 Association of CarerQol-VAS and SRB

Table 2.3 presents models to explain CarerQol-VAS and SRB, both on the basis of the CarerQol-7D scores. The CarerQol-VAS model shows that problems with mental and physical health and with performing daily activities were negatively associated with the CarerQol-VAS, and fulfilment positively. This model accounted for 38% of the variance in CarerQol-VAS scores. The model explaining SRB showed similar results; mental health problems and problems with daily activities were positively related to caregiver burden, while fulfilment was negatively related to SRB and the model accounted for 38% of the variance in SRB.

Table 2.3 Results of regression analysis of CarerQol-VAS and Self-Rated Burden (SRB); standardized coefficients for CarerQol-7D, n=230

CarerQol-7D dimensions	**CarerQol-VAS**		**SRB**	
	std.coef	**p-value**	**std.coef**	**p-value**
Fulfilment	0.21	0.00	-0.22	0.00
Relational problems	-0.06	0.30	0.09	0.16
Mental health problems	-0.35	0.00	0.16	0.02
Problems with daily activities	-0.17	0.01	0.37	0.00
Financial problems	0.00	1.00	-0.01	0.83
Support	0.00	0.998	0.03	0.53
Physical health problems	-0.11	0.09	0.11	0.11
Constant	6.88		4.21	
Adjusted R^2	0.38		0.38	

2.4.5 Clinical validity

The results of the multivariate analysis between CarerQol-VAS and background characteristics are presented in Table 2.4 (first column; analyses performed in combined dataset). These results were largely in agreement with the correlation coefficients presented in Table 2.2, with the exception of the CarerQol-7D dimension financial problems. Furthermore, caring for a child, compared to caring for distant family or friends, significantly decreased CarerQol-VAS. In addition, the CarerQol-VAS was negatively related to the age of the care recipient and positively to the number of years caring.

2.4.6 Subgroups

The CarerQol-VAS of caregivers who experience little or no burden from caring, see appendix 1 for subgroup characteristics, was positively associated with fulfilment derived from caring and negatively with mental or physical health problems and problems with combining activities (analyses performed in combined dataset). In addition, a high educational level compared to a middle educational level significantly increased the score among these less burdened caregivers.

In the subgroup of caregivers with a high SRB score, the CarerQol-7D dimensions mental and physical health were also negatively associated with CarerQol-VAS scores. Furthermore, relational problems were negatively related to this score. The CarerQol-7D dimension combining activities was not associated with the CarerQol-VAS in this group, probably because most caregivers experienced these problems (not presented). CarerQol-VAS scores were also higher for those caring for their partner and those providing long-term care, while not having a paid job and intensity of care, were negatively associated with CarerQol-VAS. Lastly, a low compared to a middle educational level was positively related to this score. This same relationship was also observed among the less burdened caregivers but, probably due to the small number of respondents with a low educational level, this difference did not reach statistical significance.

The same analyses were performed for two other subgroups of caregivers; caregivers with little or no PU and caregivers with high PU. The results, not presented here, were similar and thus in support of the clinical validity of the CarerQol.

Table 2.4 Results of regression analysis of CarerQol-VAS; combined dataset, standardized coefficients; see Appendix 1 for subgroup characteristics

	All caregivers (n=315)		Caregivers with low SRB (n=83)		Caregivers with high SRB (n=145)	
	std. coef.	p-value	std. coef.	p-value	std. coef.	p-value
CarerQol-7D						
Fulfilment	0.14	0.00	0.35[a]	0.00	0.01[a]	0.90
Relational problems	-0.20	0.00	-0.13	0.21	-0.21	0.02
Mental health problems	-0.29	0.00	-0.23	0.04	-0.31	0.00
Problems with daily activities	-0.17	0.00	-0.45[a]	0.00	0.00[a]	0.95
Financial problems	0.04[a]	0.48	0.15[a]	0.17	0.08[a]	0.37
Support	-0.02	0.63	-0.05	0.65	-0.04	0.67
Physical health problems	-0.17	0.00	-0.23	0.03	-0.19	0.04
Caregiver						
Age	0.01	0.90	-0.10	0.53	0.04	0.76
Gender (ref.male)	-0.01	0.93	0.00	0.99	-0.14	0.19
Educational level (ref.lower/middle vocational)						
primary or no education	0.08	0.11	0.09	0.31	0.18	0.04
higher vocational/university	0.06	0.19	0.28	0.01	-0.06	0.45
Paid work (ref.yes)	-0.01	0.79	0.00	0.99	-0.16	0.07
Relationship with care recipient (ref.other)			[b]		[b]	
partner	0.02	0.86	0.24	0.18	0.35	0.05
parent(-in-law)	-0.03	0.76	0.10	0.48	0.23	0.34
child	-0.23	0.01	-	-	-	-
Care recipient						
Age	-0.19	0.07	0.12	0.46	-0.14	0.40
Gender (ref.female)	-0.02	0.79	0.00	0.98	-0.12	0.30
Relatively poor health of care recipient (ref. good health)	-0.05	0.25	0.03	0.77	-0.08	0.31
Continuous surveillance (ref.no)	0.00	0.95	-0.07	0.50	-0.03	0.69
Care situation						
Years of caregiving	0.08	0.07	0.14	0.15	0.14	0.09
Days p/wk caregiving	-0.09	0.13	-0.13	0.34	-0.22	0.05
Hours p/wk caregiving	-0.04	0.50	-0.09	0.49	-0.05	0.58
Use of professional home care (ref.no)	0.03	0.51	0.06	0.53	0.03	0.77
Use of day care (ref.no)	0.03	0.59	0.00	0.97	0.11	0.17
Care recipient shares household (ref.no)	-0.02	0.83	0.09	0.68	0.11	0.45
Constant	9.19		7.32		8.80	
Adjusted R^2	0.43		0.45		0.24	

[a] Categories 'some' and 'a lot' (or 'no' and 'some' in case of fulfilment) are combined due to a small number of respondents. [b] Reference category is 'other' or 'child' due to a small number of respondents.

2.5 Discussion

There is increasing consensus that economic evaluations of health care interventions should include the impact on informal caregivers whenever informal care potentially plays an important role in order to arrive at optimal policy decisions. The CarerQol is intended to measure and value those caregiver outcomes. This study investigated the convergent and clinical validity of this instrument in a heterogeneous population of caregivers.

2.5.1 Convergent validity

As expected, the CarerQol-VAS was negatively associated with SRB and the negative dimensions of the CarerQol-7D, and positively with PU and the positive dimensions of the CarerQol-7D. Most of these correlations had moderate strength albeit the dimensions relational problems, financial problems and support had low convergent validity and the support dimension was not statistically significantly related to CarerQol-VAS. We also found evidence for the validity of the CarerQol-7D. The positive items of CarerQol-7D were positively associated to PU and negatively to SRB, while the reverse was true for the negative items of CarerQol-7D. These results are nearly identical to those reported by Brouwer et al. (2006). Overall, the results of this study suggest that the CarerQol shows moderate, but not unsatisfactory, convergent validity, given the broad outcome measure of happiness. Still, the CarerQol-7D explained the variation in CarerQol-VAS and the narrower concept of burden measured with SRB more or less equally well and both were not fully explained by it (explained variance 38%). It would be worthwhile to investigate whether a more targeted valuation component, for instance measuring ‘care-related quality of life’ – in analogy to health-related quality of life, could be meaningfully formulated and useful in this context to avoid too much influence of non-caregiving related variables.

Initially, the analyses to test the convergent validity were performed using both parametric and non-parametric tests (not presented) in order to adjust for the fact that, although VAS scores are generally considered as interval variables, they must be regarded as ordinal (e.g., [129]). The results of these tests are comparable, legitimizing the use of parametric tests in this study. This finding is confirmed by research on the methodological assumptions of the concept of happiness [124].

2.5.2 Clinical validity

The CarerQol-VAS reflects differences in important background characteristics of informal care; the relationship between caregiver and care recipient, age of care recipients and the duration of caregiving were significantly associated with CarerQol-VAS. Brouwer et al. (2006) showed comparable results. In addition, they found other factors related to happiness: partner and health status of the caregiver and the use of day care by the care recipient. However, their model differed somewhat from the model used in this study. Using a model resembling theirs the results become more similar. For example, the health of the caregiver was also positively associated with CarerQol-VAS (results not shown).

The dimensions of CarerQol-7D significantly explained the difference in CarerQol-VAS scores among subgroups of caregivers. Specifically, CarerQol-VAS was positively associated with fulfilment among caregivers with low subjective burden and negatively with relational problems among those with a high subjective burden.

2.5.3 Limitations of the CarerQol

An important limitation of the CarerQol is the difficulty of including its results in economic evaluations. The CarerQol outcomes cannot be summed with patient outcomes, however, can be included next to patient effects in cost-consequence analyses or as a separate item in a multi-criteria analysis. Standard (utility) scores for 'care profiles' defined by the CarerQol-7D are not (yet) available, but would facilitate the use of the CarerQol in economic evaluations. Future research will focus on deriving such utility scores, i.e., 'tariffs', potentially using care-related quality of life as relevant measure, in analogy to the tariffs available for the EuroQol-5D [130, 131].
In addition, the non-significant relation of the support dimension of the CarerQol-7D deserves attention. This non-significant relation may be due to a lack of power, but may also imply that support is less relevant in this context, that it measures different aspects, e.g., professional support or informal support, or that support is already indirectly captured in other CarerQol-7D dimensions. This issue of content validity deserves attention in future research.

This study, like the first test [110], was cross-sectional. Especially in the context of economic evaluations, it is important to test the sensitivity of the instrument to changes in the caregiving situation, which requires a longitudinal study set-up. Moreover, the reliability of the CarerQol instrument needs to be investigated as well.

2.5.4 Limitations of the study design

Some limitations of this study deserve mentioning. First, the results cannot be straightforwardly generalized, because the study sample is not necessarily representative of the population of Dutch informal caregivers. Respondents were contacted via regional support centres and therefore are expected to be relatively burdened [132]. Indeed, the mean SRB score of 5.2 and the mean CarerQol-VAS score of 6.3 indicate that although caregivers in the study sample were fairly happy, they still experienced substantial caregiving burden. As a reference, the mean SRB in a population of caregivers of stroke patients was 3.0 [11], and 5.8 in a heterogeneous sample of Dutch caregivers, very similar to the current sample [110]. Moreover, the average happiness of the general population in the Netherlands, measured on a comparable scale, was 7.8 [133]. The relatively high burden among caregivers in the study sample may have influenced the results. Nevertheless, for the purpose of this study, which was testing the psychometric properties of the CarerQol, such a bias seems unproblematic. In addition, the external validity may be relatively low, because the non-response rate was relatively high. Possible reasons for a relatively high non-response are lack of time or energy to complete the questionnaire among caregivers due to the high strain placed on them by the care situation. Additionally, some informal caregivers may not regard their activities as 'informal care' and therefore do not feel the need to participate in research on this subject. To overcome these biases, the convergent and clinical validity of the CarerQol should also be tested in samples of caregivers not selected via support centres. It may also be worthwhile to test the validity among a specific population of caregivers, e.g., caregivers of persons with a specific disease.

2.6 Conclusions

Our study confirmed results of the first test of the convergent and clinical validity of the CarerQol. Overall, the CarerQol provides a good description of the impact of caregiving on informal caregivers and therefore can be seen as a useful instrument to include this effect in economic evaluations. To improve its use(fullness) to measure and value the effects of caregiving, future research should further investigate its content validity, reliability, and sensitivity and the development of utility scores for the 'care profiles'. Ultimately, the aim is to adequately inform policy makers about the effects of interventions on caregivers and, as such, support informed decisions in the field of health care.

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Appendix 1

Table A2.1 Percentages or mean (sd) of variables for subgroups in Table 2.4.

	Caregivers with low SRB (n=114)	Caregivers with high SRB (n=165)
CarerQol-7D		
Fulfillment no or some	32.5%	60.6%
a lot	67.5%	39.4%
Relational problems no	64.9%	21.2%
some	28.1%	44.2%
a lot	7.0%	34.6%
Mental health problems no	55.3%	17.0%
some	39.5%	50.3%
a lot	5.3%	32.7%
Problems with daily activities no	51.8%	8.5%
some or a lot	48.3%	91.5%
Financial problems no	86.0%	70.3%
some or a lot	14.0%	29.7%
Support no	23.9%	28.5%
some	50.9%	57.6%
a lot	25.4%	13.9%
Physical health problems no	55.3%	16.4%
some	36.8%	49.7%
a lot	7.9%	33.9%
Caregiver		
Age	56.99 (12.85)	59.40 (12.67)
Gender female	76.3%	72.7%
male	23.7%	27.3%
Educational level primary or no	5.3%	14.1%
lower/middle vocational	73.5%	55.8%
higher vocational/university	21.2%	30.1%
Paid work yes	35.4%	28.7%
no	64.6%	71.3%
Relationship with care recipient partner	38.4%	52.2%
parent(-in-law)	34.8%	33.7%
child or other	27.0%	14.1%
Care recipient		
Age	69.91 (19.17)	68.63 (20.50)
Gender female	56.6%	52.1%
male	43.4%	47.9%
Health (rated by caregiver) relatively good	45.5%	29.9%
relatively poor	54.5%	70.1%
Surveillance 24/7 yes	23.2%	40.5%
no	76.8%	59.5%
Care situation		
Years care giving	5.98 (5.43)	9.38 (9.63)
Days p/wk care giving	5.54 (2.30)	5.98 (1.93)
Hours p/wk care giving	30.20 (31.63)	47 (43.75)
Care recipient shares household yes	48.3%	67.3%
no	51.8%	32.7%
Professional home care yes	51.3%	66.7%
no	48.7%	33.3%
Day care yes	16.1%	33.3%
no	83.9%	66.7%

Note: Number of respondents differs from Table 2.4, because in Table 2.4 only cases without missing values on the dependent variables in the regression analysis were included.

CHAPTER

3

Sustained informal care: The feasibility, construct validity and test–retest reliability of the CarerQol instrument to measure the impact of informal care in long-term care

Based on Hoefman, R. J., van Exel, N. J. A., Foets, M., & Brouwer, W. B. (2011). Sustained informal care: The feasibility, construct validity and test–retest reliability of the CarerQol instrument to measure the impact of informal care in long-term care. Aging & Mental Health, 15(8), 1018-1027.

Abstract

Objectives

This study analyses feasibility, construct validity and test-retest reliability of the CarerQol instrument among informal caregivers of long-term care (LTC) users. The CarerQol measures the impact of informal care by assessing happiness (CarerQol-VAS) and describing burden dimensions (CarerQol-7D).

Methods

Data was gathered among informal caregivers of patients obtaining day care or living in a LTC facility in the Netherlands with two questionnaires sent with a two-week interval (n test=108, n retest=100). Percentages of missing values indicated feasibility of the CarerQol. Construct validity assessed the extent to which differences in background characteristics were reflected in happiness scores (CarerQol-VAS) with bivariate and multivariate statistics. Additionally, construct validity was tested with assessing the correlation between the CarerQol and the Self-Rated Burden scale (SRB). Percentages of complete agreement between CarerQol scores at test and retest, Cohen's kappa coefficients (κ value) and Intraclass Correlation Coefficients (ICC) were used to assess reliability.

Results

3.7% was missing on the CarerQol. CarerQol-VAS was positively associated with caregivers' age and health, and negatively with SRB score. The percentage of complete agreement of CarerQol-VAS between the measure moments was 60% and between 76% and 96% for CarerQol-7D. κ value and ICC of CarerQol-VAS were 0.52 and 0.86, and ranged between 0.55 and 0.94 for CarerQol-7D.

Conclusion

The CarerQol measures the impact of informal care among caregivers of LTC users in a feasible, valid and reliable way.

3.1 Introduction

In the Netherlands, about one in five persons provide care to an ill or disabled family member or friend [1], usually in the home of the care recipient [14]. Although this is often the preferred option [28] additional professional care may be needed. Using professional care is often a gradual process; from professional home care, to the option of day care in a long-term care (LTC) facility ending with the final alternative of admission to a nursing home [134]. After institutionalization many care recipients will still receive care from their family or friends, and the care provided by these caregivers plays an important role in LTC facilities [135, 136]. The need for this involvement of informal care is likely to increase in the future due to a further scarcity of professional LTC, caused by an expected increase in the demand of nursing home care due to the ageing population [137] and a shortage of health care personnel in the labour market [60].

Informal caregivers perform different tasks in LTC facilities, such as doing laundry, assisting with meals, and keeping company [135, 138-140], which may well be rewarding for informal caregivers. This informal care may reduce the workload of formal caregivers, which is desirable in light of the anticipated shortage of formal care. Nevertheless, this substitution is unsustainable when it imposes too much burden on informal caregivers. Most caregivers of LTC residents experienced high burden before institutionalization of the care recipient. This burden seemed to be a risk factor for institutionalization [141-143]. However, whether institutionalization influences this burden remains unclear [144, 145]. The level of burden among caregivers of residential and institutionalized patients is sometimes shown to be comparable [138], while in other occasions, institutionalization relieves the physical [146, 147] and psychological burden of caregivers [144, 146, 147]. Others found that LTC placement does not improve the emotional health of caregivers [142, 148], not even after a longer period of residence [142, 149, 150]. Over time, some types of emotional outcomes may improve, like anxiety [142], depressive symptoms [146], and mental health [149]. In addition, duration of institutionalization seems positively related to positive changes in serious medical symptoms in informal caregivers [146]. These diverse findings may indicate that after institutionalization caregivers experience divergent effects. On the one hand, the burden of care will diminish, due to substitution of tasks to formal care. On the other hand, informal caregivers may experience more burden related to the physical separation from the patient. Moreover, institutionalization is often associated with deteriorated health of the patient. Consequently, the family effect, i.e., the direct influence of the health status of patients on the well-being and health of their family and friends arising from 'the fact that we care about other people and their health' [26, 53] will increase. Furthermore, caregivers of institutionalized patients encounter new potential sources of stress, e.g., communication with professional caregivers, handing over the patient's care, while still maintaining a level of control over it [151] and interaction with the families of other residents [136]. Moreover, caregivers may experience feelings of guilt related to the institutionalization decision [152].

A feasible, valid and reliable instrument is needed to measure the burden imposed on caregivers of LTC users. Research on the impact of informal care often measures this in terms of objective [91] or subjective burden [33, 106, 107, 121], impact on health and general quality of life or well-being of the caregiver [25, 26, 50].

3.1.1 Study objectives

The CarerQol (Care-related Quality of Life) instrument [110] consists of a subjective burden measure and an assessment of overall well-being. The CarerQol measures the individual experience of the informal care situation and combines the information density of a subjective burden measure with an overall assessment of the impact of informal care.

This chapter addresses whether the CarerQol is a feasible, valid and reliable instrument to assess the burden among caregivers of LTC users. Previous studies analysed feasibility and construct validity of the instrument among caregivers providing informal care at home ([110] and chapter 2), but did not address the reliability of the CarerQol instrument. Moreover, no information is available on the psychometric properties of the instrument among caregivers of LTC users.

3.2 Methods

3.2.1 Data

Data have been gathered with written questionnaires mailed by post to 319 contact persons of patients using care provided by a nursing home near Rotterdam, the Netherlands, in May and June 2009. The gross response rate at the first measurement moment (*t0*) was 39.2%. Respondents were asked whether they 'provided support or care to a person living in the LTC facility or using day care in this facility'. Those answering negatively were excluded, leaving a response rate of 33.9% (n=108). The care provided by this LTC facility consists of two types: nursing care services for permanent residents of a somatic or psycho-geriatric ward and day care. In the study sample, 74% of the respondents provided informal care to institutionalized patients and 26% to patients using day care (Table 3.1).

In order to study the test-retest reliability, a second questionnaire (retest) was sent to all respondents two weeks after the first, irrespectively of participation in the first survey. A cover letter explained the study purpose and offered a small donation to a fund for patients of the LTC facility if both questionnaires would be returned. The net response rate of the second questionnaire (*t1*) was somewhat lower: 31.3% (n=100). The response rate is moderate, but still higher than achieved in other studies among informal caregivers [28, 105].

3.2.2 Questionnaire

The impact of informal care was measured with the CarerQol instrument [110], the Self-Rated Burden scale (SRB; [105]), and the Assessment Scale of the Informal caregiving Situation (ASIS). The CarerQol instrument describes caregiver burden on seven dimensions (CarerQol-7D) and valuates general quality of life (CarerQol-VAS) (see Figure 3.1). The CarerQol-7D is composed of two positive dimensions of care: fulfilment and support, and five negative dimensions: relational problems, mental health problems, problems with combining daily activities, financial problems and physical health problems, with answering categories 'no' (i), 'some' (ii), and 'a lot of' (iii). The CarerQol-VAS measures happiness: 'the degree to which an individual judges the overall quality of his life-as-a-whole favourably' [125] with a horizontal Visual Analogue Scale (VAS) with 0, 'completely unhappy', and 10, 'completely happy', as endpoints [110]. The SRB measures the overall burden of informal care as perceived by the carer with a horizontal VAS ranging from 'not straining at all' (0) to 'much too straining' (10) [105]). The ASIS, newly introduced here, consists of a horizontal VAS with endpoints 'worst imaginable caregiving situation' (0) and 'best imaginable caregiving situation' (10), coinciding with the two extreme CarerQol-7D profiles: a lot of problems and no fulfilment or support, and no problems and a lot of fulfilment and support. We will study whether the CarerQol instrument could be extended with this scale, because the CarerQol-VAS appears to be a fairly broad outcome measure which may be less sensitive to non-substantial changes in the care situation. In this way, the ASIS could serve as a valuation component of informal care which is less influenced by non-caregiving factors.

Further, the questionnaire included questions on background characteristics of the caregiver, care recipient and caregiving situation. Specifically, information was obtained on the caregiver's age, gender, having a partner, educational level, vocation, relation to care recipient, and health status and on the care recipients' age, gender, health, and level of independence. The level of independence on bathing, dressing, toileting, transferring, continence, and feeding was measured with the KATZ Index of Independence in Activities of Daily Living [153]. Questions on the caregiving situation included duration of care (in years), intensity of care (in days per week and hours per day), caregiving tasks divided up in activities of daily living (ADL), instrumental activities of daily living (IADL), personal care and questions on visiting the care recipient, institutionalization and need for permanent surveillance of the care recipient, and whether other informal caregivers provided support.

The second questionnaire (retest) contained the CarerQol instrument and questions to detect differences in the health status of the caregiver and care recipient, and differences in the caregiving situation during the two-week interval. These questions were included to ensure that the CarerQol scores at *t0* and *t1* were comparable and useful for assessing test-retest reliability and not influenced by significant changes in the caregiving situation during the past two weeks.

3.2.3 Statistical analyses

Descriptive statistics of the study sample were calculated. The feasibility of the CarerQol instrument was assessed using percentages of respondents with missing values ([105, 110] and chapter 2). Missing values were computed for the complete CarerQol instrument, the CarerQol-VAS, the CarerQol-7D, and for separate CarerQol-7D dimensions.

In order to study the construct validity of the CarerQol instrument, respondents with missing values on the CarerQol have been excluded (n=4). Construct validity was assessed in two ways: clinical and convergent validation. We refer to clinical validation as the extent to which differences in background characteristics, such as caregiver variables, are reflected in CarerQol-VAS scores. This was studied by the association between CarerQol-VAS and background characteristics with One-way ANOVA tests for categorical variables, and Spearman's correlation coefficients for ordinal or interval variables. In addition, a stepwise multiple regression analysis (backward selection, $p<0.2$) of CarerQol-VAS and CarerQol-7D and background characteristics was performed. The choice of the reference category of categorical variables in this model was based on the highest mean CarerQol-VAS score. Some categories of these variables have been merged due to a small number of cases in one of these categories (<10% of observations). The items of the CarerQol-7D are continuous in the model and were included despite of their level of statistical significance. Convergent validation assesses whether the underlying construct of the CarerQol instrument resembles the construct of the other subjective burden measure: SRB, with the use of Spearman's correlation coefficients. In this study, the strength of the Spearman's correlation coefficients is indicated by the guideline of Hopkins (2002): <0.1 trivial; 0.1-0.3 small; 0.3-0.5 moderate; 0.5-0.7 high; 0.7-0.9 very high; >0.9 nearly perfect, and by comparison with previous studies ([110] and chapter 2). We consider correlations of small to moderate strength or higher as a sign of validity, because happiness is a broad outcome measure and may not only relate to caregiving, but might also be influenced by other aspects of life, and by the care recipient's bad health condition ('family-effect', [26, 53]).

For test-retest reliability analyses, respondents who did no longer provide informal care at *t1* were excluded (n=7). We also disregarded respondents who indicated that their or the care recipients' health status or the caregiving situation considerably changed during the period of study (n=5). The test-retest reliability of the CarerQol instrument was assessed with percentages of complete agreement, Cohen's kappa coefficients (κ value) and Intraclass Correlation Coefficients (ICCs). Percentages of complete agreement indicate the proportion of cases with the same value on the CarerQol instrument at *t0* and *t1*. To adjust for the fact that a number of these agreements may arise by chance alone, kappa coefficients that produce chance-corrected agreements were also used. ICCs were presented because the CarerQol instrument consists of scaled responses and therefore not only total agreement, but also partial agreement is of importance [98], e.g., the small difference between the CarerQol-VAS of 7.0 at *t0* and 6.5 at *t1*. Different ICCs can be used depending on the type of analysis of variance needed for the study design, whether the differences in scores between persons are relevant and the unit of analysis (individual or mean score) [154]. This study focuses on the difference in scores by single respondents on different measurement moments. Whether person A rates his/her happiness higher than person B is irrelevant in this context. In addition, self-rated instruments were studied and therefore no 'external judges' were used, in contrast to for example studies evaluating medical diagnostic instruments. Based on these arguments, ICC(1,1) was used, a Case 1 ICC using a One-way ANOVA with a random error component to analyse individual scores. Confidence intervals of k values and ICCs were used to assess the size of the difference between the scores on *t0* and *t1*. In this study, the guideline of Altman (1991) was used indicating the strength of agreement: κ value < 0.20 poor; 0.21-0.40 fair; 0.41-0.60 moderate; 0.61-0.80 good; 0.81-1.00 very good [155]. To further assess the stability of the CarerQol-VAS over time, differences in CarerQol-VAS at *t0* and *t1* were analysed and a regression analysis of CarerQol-VAS at *t0* and *t1* was performed.

Statistical significance was based on an alpha error of 0.05. All statistical analyses were performed with Stata® version 11.0 (StataCorp LP).

3.3 Results

Table 3.1 presents a description of the caregivers, care recipients and care situations in the study sample. The subjective burden of caregiving was moderate (mean SRB score of 4.9), but far from 'best imaginable' (mean score of 6.7 on ASIS). The problem most often reported was combining care with other daily activities (66.7%, Figure 3.1). Relational problems with the care recipient (54.7%) and physical health problems (54.6%) were also experienced by many caregivers; most caregivers indicated to have at least some problems on these CarerQol-7D dimensions. A problem also encountered by a relatively large group of caregivers (43.9%), although not by the majority, was mental health problems. Overall, the mean CarerQol-VAS score was 7.0

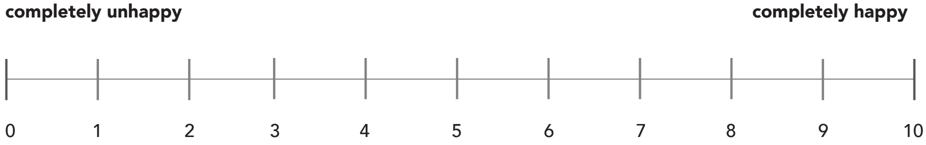
We would like to form an impression of your caregiving situation.
Please tick a box to indicate which description best fits your caregiving situation at the moment.

Please tick only one box per description: 'no', 'some' or 'a lot of'.

	no	**some**	**a lot of**	
I have	8.5%	44.3%	47.2%	fulfilment from carrying out my care tasks.
I have	45.3%	41.5%	13.2%	relational problems with the care receiver (*e.g., he/she is very demanding or he/she behaves differently; we have communication problems*).
I have	56.1%	37.4%	6.5%	problems with my own mental health (*e.g., stress, fear, gloominess, depression, concern about the future*).
I have	33.3%	56.5%	10.2%	problems combining my care tasks with my daily activities (*e.g., household activities, work, study, family and leisure activities*).
I have	87.9%	11.2%	0.9%	financial problems because of my care tasks.
I have	23.6%	55.7%	20.8%	support with carrying out my care tasks, when I need it (*e.g., from family, friends, neighbours, acquaintances*).
I have	45.4%	47.2%	7.4%	problems with my own physical health (*e.g., more often sick, tiredness, physical stress*).

How happy do you feel at the moment?

Please place a mark on the scale below that indicates how happy you feel at the moment.



Note: Observed score in study sample (mean (SD)): 7.0 (1.6)

Figure 3.1 CarerQol instrument

3.3.1 Feasibility

A small percentage of respondents in the study (3.7%) had one or more missing values on items of the CarerQol instrument (not presented in table). Less than one per cent of the respondents did not answer the CarerQol-VAS, while approximately three per cent did not answer at least one of the CarerQol-7D dimensions.

3.3.2 Clinical validation

Bivariate analysis of CarerQol-VAS and background characteristics showed that happiness was positively correlated with caregiver's health status. Further, happiness was higher among caregivers who provide care to their parents (as compared to those who care for their partner or others), to older and female care recipients, and when the caregiving task was less intensive (in days per week and hours per day). In addition, the CarerQol-VAS score was higher when the care recipient was institutionalized and in case caregivers visited these care recipients less frequently (Table 3.2). Multivariate stepwise regression analysis showed that, in addition to the CarerQol-7D dimensions, caregivers' age and health, and whether caregivers had a partner (C.I. 0.90) were significant associated variables of happiness. Note that the sign of the coefficient of institutionalization reversed in the multivariate model, although not statistically significant (Table 3.2).

Table 3.1 Characteristics of the sample at t*0* (mean (SD) or in percentages), (n=108)

		mean (SD)	%
Caregiver			
Age		59.1 (11.0)	
Gender	female		71.4
	male		28.6
Partner	yes		82.7
	no		17.3
Educational level	primary (or lower)		16.5
	lower/middle vocational		56.7
	higher vocational/university		29.8
Paid work	full-time		23.1
	part-time		30.1
	no		46.2
Subjective health (0-10)		7.2 (1.5)	
Care recipient			
Age		79.6 (12.9)	
Gender	female		69.5
	male		30.5
Health (rated by caregiver; 0-10)		4.7 (1.9)	
KATZ (ADL independence; 0-6)		2.4 (2.0)	
Care situation			
Relationship with care recipient	partner		22.9
	parent (-in-law)		57.1
	other		20.0
Duration of caregiving (years)		5.7 (6.6)	
Intensity of caregiving	days/wk	3.0 (2.6)	
	hours/wk	16.4 (30.6)	
Care activities (h/wk)	adl-activities	5.5 (11.6)	
	iadl-activities	6.2 (13.8)	
	personal care	5.6 (19.7)	
Visiting care recipient (h/wk) [a]		9.5 (8.5)	
Care recipient is institutionalized (yes)			74.0
Support from other caregivers (yes)			56.2
Permanent surveillance [b] (yes)			35.0
SRB (0-10)		4.9 (2.2)	
ASIS (0-10)		6.7 (1.7)	

[a] Caregivers of institutionalized persons only; [b] Caregivers of day care users only.

Table 3.2 Bivariate (n=104; Spearman's rho; 2-tailed) and multivariate (n=84, stepwise regression analysis) analyses of CarerQol-VAS with characteristics of the caregiver, the care recipient and the care situation

		mean CarerQol-VAS	**p-value [f]**	**Spearman's rho**	**p-value**	**std. coef.**	**p-value**
Caregiver							
Age	<59 years	7.12	0.48	-0.12	0.24	-0.23	0.02
	≥59 years	6.89					
Gender	female	6.90	0.28	-	-	-	-
	male	7.28					
Partner	yes	7.05	0.69	-	-		
	no	6.88				-0.17 [d]	0.08
Educational level	primary (or lower)	6.83	0.93	0.05	0.74	-	-
	lower/middle vocational	7.01				-	-
	higher vocational/ university	7.05				-	-
Paid work	full-time	6.96	1.00	0.06	0.54	-	-
	part-time	6.98				-	-
	no	7.05				-	-
Subjective health (0-10)	<7	6.49	0.00	0.42	0.00	0.35	0.00
	≥7	7.55					
Care recipient							
Age	<80 years	6.44	0.07	0.23	0.02	0.15	0.14
	≥80 years	7.31					
Gender	female	7.18	0.04	-	-	-	-
	male	6.48				-	-
Health (rated by caregiver; 0-10)	<5	6.89	0.66	-0.03	0.80	-	-
	≥5	7.03				-	-
ADL independence (0-6)	<2.5	7.06	0.70	0.06	0.53	-	-
	≥2.5	6.93					
Care situation							
Relationship with care recipient	partner	6.13	0.01	-	-	-	-
	parent (-in-law)	7.29				-	-
	other	7.03				-	-
Duration of caregiving	<6 years	6.97	0.67	0.06	0.53	-	-
	≥6 years	7.12				-	-
Intensity of caregiving	<3 days/wk	7.43	0.00	-0.29	0.00	-	-
	≥3 days/wk	6.51				-	-
	<14.5 h/wk	7.18	0.08	-0.27	0.01	-	-
	≥14.5 h/wk	6.50				-	-
Care activities							
adl-activities	<5 h/wk	7.26	0.01	-0.13	0.24	-	-
	≥5 h/wk	6.22				-	-
iadl-activities	<4 h/wk	7.09	0.63	-0.25	0.01	-	-
	≥4 h/wk	6.93				-	-
personal care	<6 h/wk	7.19	0.24	-0.30	0.00	-	-
	≥6 h/wk	6.68				-	-
Visiting care recipient [a]	<9 h/wk	7.13	0.60	-0.27	0.02	-	-
	≥9 h/wk	6.95				-	-
Care recipient is institutionalized	yes	7.13	0.05 [c]	-	-		
	no	6.43				0.13 [e]	0.17
Support from other caregivers	yes	7.22	0.15	-	-	-	-
	no	6.75				-	-
Permanent surveillance [b]	yes	7.27	0.38	-	-	-	-
	no	6.50				-	-

Note: Stepwise multivariate regression analysis is also corrected for CarerQol-7D dimensions. Age of caregiver, subjective health of caregiver and age of care recipient are continuous variables and partner of caregiver and institutionalization of care recipient are dummy variables in the stepwise regression model. [a] Caregivers of institutionalized persons only; [b] Caregivers of day care users only; [c] p=0.049; [d] Reference category of variable 'partner' in stepwise regression analysis is 'yes, partner'; [e] Reference category of variable 'care recipient is institutionalized' in stepwise regression analysis is 'yes, institutionalized'; [f] One-way ANOVA test

3.3.3 Convergent validation

Table 3.3 shows the bivariate correlations between CarerQol-VAS, CarerQol-7D, SRB and ASIS. CarerQol-VAS was positively associated with the positive CarerQol-7D dimensions fulfilment and support, however not statistically significantly (range absolute values correlation coefficients 0.05-0.15). A negative association was observed between CarerQol-VAS and the negative CarerQol-7D dimensions (range absolute values correlation coefficients 0.26-0.58). In addition, a negative association was observed with SRB and a positive association with ASIS.

The statistically significant associations between the dimensions of the CarerQol-7D and SRB and ASIS were all in the expected directions (range absolute values correlation coefficients 0.23-0.44). Combining daily activities and mental and physical health problems were statistically significantly and positively associated with SRB. Further, relational problems, combining daily activities, and mental and physical health problems were negatively and fulfilment was positively and statistically significantly associated with ASIS.

Table 3.3 Bivariate correlation analysis of CarerQol-VAS, CarerQol-7D, SRB and ASIS at t*0*, (Spearman's rho; 2-tailed)

	CarerQol-VAS		**SRB**		**ASIS**	
	n=104	**p-value**	**n=102**	**p-value**	**n=101**	**p-value**
CarerQol-7D						
Fulfilment	0.15	0.14	0.00	1.00	0.27	0.01
Relational problems	-0.26	0.01	0.15	0.13	-0.23	0.02
Mental health problems	-0.58	0.00	0.36	0.00	-0.37	0.00
Combining daily activities	-0.32	0.00	0.44	0.00	-0.34	0.00
Financial problems	-0.30	0.00	0.05	0.59	-0.04	0.71
Support	0.05	0.60	-0.06	0.56	0.13	0.19
Physical health problems	-0.44	0.00	0.38	0.00	-0.41	0.00
SRB	-0.33	0.00				
ASIS	0.47	0.00	-0.34	0.00		

3.3.4 Test-retest reliability

The test-retest reliability statistics are presented in Table 3.4. The CarerQol-VAS scores were very similar at *t0* and *t1* (2 week interval); the mean difference was 0.04 (standard deviation 0.64, not presented). Bivariate regression of CarerQol-VAS at *t0* and *t1* showed high correlation (0.96; p<0.001, not presented). Table 3.4 shows that 60 per cent of all cases reported the same CarerQol-VAS score at *t0* and *t1*. The κ value and ICC of the CarerQol-VAS were 0.52 and 0.86. The percentage of complete agreement of the CarerQol-7D was between 76 and 96 per cent. The κ values of the CarerQol-7D ranged between 0.59 and 0.81. ICCs of the CarerQol-7D had values between 0.55 and 0.94.

Table 3.4 Test-retest reliability of CarerQol instrument in percentages of complete agreement, kappa statistics and Intraclass-correlation coefficients (ICC) among caregivers with a comparable health status and caregiving situation at t*0* and t*1* (2-week interval)

	Percentage complete agreement	κ value	95% CI κ value		ICC	95% CI ICC value	
			Lower bound	Upper bound		Lower bound	Upper bound
CarerQol-VAS[c]	60	0.52	0.40	0.65	0.86	0.71	1.01
CarerQol-7D							
Fulfilment [a]	84	0.72	0.58	0.86	0.75	0.33	1.18
Relational problems [a]	76	0.60	0.44	0.76	0.69	0.20	1.18
Mental health problems [b]	78	0.59	0.42	0.75	0.55	0.00	1.17
Combining daily activities [d]	88	0.79	0.66	0.91	0.81	0.44	1.17
Financial problems [c]	96	0.81	0.57	1.00	0.94	0.79	1.09
Support [a]	79	0.65	0.50	0.80	0.70	0.27	1.14
- Physical health problems [c]	88	0.78	0.65	0.91	0.80	0.41	1.19

[a] n=80; [b] n=81; [c] n=82; [d] n=83

3.4 Discussion

This study shows that the CarerQol instrument is a feasible instrument and measures burden among caregivers of LTC users in a valid and reliable way. More specifically, the CarerQol has excellent feasibility, given that almost all caregivers completed all questions of the instrument. Concerning clinical validation, differences in CarerQol-VAS scores were found in relevant subgroups of caregivers. Especially, caregivers' characteristics influenced the happiness among caregivers of LTC users. Specifically, health status and age of the caregiver and having a partner were related to happiness. A possible explanation for the importance of these caregiver characteristics is that the care recipients in this sample largely possessed the same characteristics, such as female sex and relatively bad health. In addition, in all caregiving situations the majority of the care was provided by professional caregivers. As was the case in this study, previous studies confirm the relatively modest role of care recipient characteristics on caregiver burden in the context of LTC care [145, 156]. As with happiness, the factors related to burden of caregivers of LTC users were largely caregiver and care situation characteristics, such as gender [145], age, involvement in caregiving, perceptions of the quality of care [145, 156], and duration of informal care prior to institutionalization [156]. Obviously, the fact that the study sample was relatively small and selective warrants caution in interpreting and generalizing the results.

The results of convergent validation show that the associations of the CarerQol with the other measure of caregiving burden were as expected and had moderate strength. For example, CarerQol-VAS was negatively related to negative items of CarerQol-7D and subjective burden (SRB), and positively to positive items of CarerQol-7D. In addition, a higher reported burden on the CarerQol-7D was associated with higher SRB. Previous studies on the psychometric properties of the CarerQol in two heterogeneous samples of informal caregivers have also shown that the feasibility [110], clinical and convergent validity statistics were moderate to good ([110] and chapter 2). These studies reported a low number of missing items on the CarerQol instrument, stated that the CarerQol discriminated well between groups of caregivers, and that the associations between the CarerQol and other burden measures were in the expected direction.

This study presents the first results concerning the reliability of the CarerQol instrument. When considering the consistency of the responses the CarerQol-VAS had good test-retest statistics. This conclusion is supported by other results. First, the happiness score at the second measurement moment was importantly explained by the happiness score at the first measurement moment. Furthermore, the mean difference between these two CarerQol-VAS scores was nearly zero. Overall, the test-retest statistics of the other part of the instrument, CarerQol-7D, were also good. For the dimension financial problems very good test-retest statistics were found, which is likely to be related to the short follow-up period and the large number of respondents indicating ‘no problems’ (87.5% of sample). The dimension mental health problems had moderate test-retest statistics, which may require future attention. This may to some extent be explained by the fact that especially this variable may fluctuate most within a two week period.

In addition, this study shows, in contrast to the prevailing belief that institutionalization implies that informal caregivers simply hand over all care tasks to LTC institutions and subsequently are no longer involved in caregiving, that even after institutionalization informal caregivers of LTC users experienced a considerable burden of caregiving, both in terms of time and subjective burden. The data of this study indicates that despite the continued involvement in caregiving, even after institutionalization, the well-being of caregivers is relatively high, although some aspects of burden remain prominent. Especially, combining daily activities with care was an important problem when caring for LTC users. The majority of the study sample was middle-aged or older, provided care to their parents, and had a paid job. Consequently, these caregivers have to provide care next to performing paid work. In addition, they first have to travel to their parents’ (nursing) home, while normal (caring) tasks in their household need to be performed as well. It is not surprising, therefore, that the different roles as employee, parent, partner, and caregiver, may lead to role conflict and time pressure.

Moreover, caregivers of LTC users often had relational problems with the care recipient and reported own health problems. The presence of relational problems can be partly explained by the relatively large proportion of care recipients with mental health problems, e.g., Alzheimer’s disease, because the study sample included a psycho-geriatric ward. Information on experienced problems, also in light of the significant time investment of on average 16 hours per week, can be used to monitor the position of caregivers and help them in a sustained involvement in the care for their loved ones. This is important to note in times when increased pressure on formal care may result in more demand for informal care.

3.4.1 Study limitations

Our sample of caregivers was of modest size, contained caregivers of one LTC institution and (deliberately) consisted only of caregivers of LTC users. As a consequence, the results of this study, especially the results on reliability which are only valid for this sample of caregivers, should be interpreted with caution. Therefore, it could be worthwhile to perform a similar test-retest experiment in a heterogeneous sample of caregivers.

The small number of respondents restricted the analysis of subgroups. In future studies using larger samples, it would be interesting to perform subgroup analyses studying subjective burden of caregivers of institutionalized persons, and distinguishing among them on the basis of duration of stay, and of persons only receiving day care and still living in their own homes. Such studies could also further investigate test-retest reliability in subgroups.

Another limitation of the study is that, although the retest questionnaire was sent out two weeks after the test questionnaire, the indicated dates of completing these questionnaires revealed that this time-interval was sometimes shorter or longer than two weeks. For example, 23 respondents filled in both questionnaires within one week while three respondents had a time interval of more than three weeks. Additional analyses, however, showed that the mean difference in the CarerQol-VAS between the two measurement moments was not significantly related to the time interval in between the two measurement moments.

The statistical analysis used for clinical validation also deserves attention. Multivariate stepwise regression analysis has been criticized [157]. One important criticism is that stepwise methods provide models that do not necessarily contain the best subset of independent variables. To avoid inaccuracy of the findings a relatively high p-value was used as the criterion to delete a variable from the model. In addition, in order to investigate the robustness of the findings, a multivariate regression model including all independent variables simultaneously was performed. This resulted in the same statistically significantly explanatory variables to emerge as in the stepwise regression, thus confirming the robustness of the findings to the method used.

Since the CarerQol instrument was used to measure the impact of informal care, some aspects of this instrument need to be discussed. First, as mentioned in the introduction, the CarerQol-VAS consists of a broad outcome measure that may be influenced by more than only the caregiving situation. Hence, the more specific VAS assessing the informal caregiving situation, ASIS, was included. This score was relatively strongly related to the CarerQol-VAS.

Secondly, coping and adaptation may affect the CarerQol-VAS scores, reducing the observed effects of caregiving on happiness. Therefore, standard (ex ante) utility scores, or tariffs, for the CarerQol instrument, like the ones available for the EuroQol instrument [130, 131], would be useful. This way, the scores on the 7D descriptive part of the CarerQol could be linked to standard utility scores. Another important issue for future research is the sensitivity of the CarerQol to changes in a longitudinal study design.

Thirdly, while the net response rate of 33.9% compares relatively favourable to previous studies that have relied on postal surveys sent to informal caregivers [28, 105], it needs noting that non-response may have been selective. For instance, caregivers experiencing either a high or a negligible burden may have been less inclined to complete the questionnaire. We could not further investigate non-response, adding to the caution of generalizing the results at this stage.

Notwithstanding the study limitations, the results suggest that the CarerQol possesses moderate to good psychometric properties, which is in line with previous research ([110] and chapter 2). Concluding, the CarerQol instrument appears to be a useful instrument for measuring the, non-negligible, impact of informal care in an often overlooked group of caregivers.

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Appendix chapter 3

Table A3.1 Results of stepwise regression analysis of CarerQol-VAS with CarerQol-7D and background characteristics of caregiver, care recipient and care situation (n=84)

	std. coef.	p-value
CarerQol-7D		
Fulfilment [a]	-0.04	0.70
Relational problems	-0.26	0.02
Mental health problems [a]	-0.33	0.00
Problems with daily activities	-0.22	0.03
Financial problems [a]	-0.13	0.16
Support	0.07	0.46
Physical health problems [a]	0.08	0.51
Age caregiver	-0.23	0.02
Health caregiver	0.35	0.00
Partner status caregiver (ref. yes)	-0.17	0.08
Age care recipient	0.15	0.14
Care recipient is institutionalized (ref. yes)	0.13	0.17
Constant	8.13	
Adjusted R^2	0.43	

[a] Reference category is 'no or some'

CHAPTER

4

Measuring the impact of caregiving on informal caregivers: a construct validation study of the CarerQol instrument

Based on Hoefman, R. J., van Exel, J., & Brouwer, W. B. (2013). Measuring the impact of caregiving on informal carers: a construct validation study of the CarerQol instrument. Health and Quality of Life Outcomes, 11(1), 173.

Abstract

Background

Informal caregivers provide a significant part of the total care needed by ill or disabled persons. Although informal care is often the preferred option of those who provide and those who receive informal care, caring can nevertheless be very straining. This study investigates construct validation of an instrument of the impact of caregiving, the CarerQol.

Methods

Data was collected among adult caregivers (n=1,244) selected from the general population using an online questionnaire in October 2010, in the Netherlands. The CarerQol measures and values the impact of informal care. The CarerQol measures subjective burden (CarerQol-7D) and well-being (CarerQol-VAS). Construct validation comprised clinical, convergent and discriminative validity tests.

Results

Clinical validity was supported by statistically significant associations of CarerQol-VAS and caregivers' health, income and employment status, care recipients' health, and the relationship between caregiver and care recipient. Convergent validity was supported by positive associations of CarerQol-VAS with the two positive CarerQol-7D dimensions (fulfilment and support) and negative associations with the five negative CarerQol-7D dimensions (relational problems, mental health problems, problems combining daily activities, financial problems and physical health problems). Moreover, CarerQol-VAS was negatively associated with other instruments measuring caregiving burden.

Conclusions

Construct validity tests in a large, heterogeneous sample of caregivers show that the CarerQol validly measures the impact of caregiving. The CarerQol can be used in informal care research and economic evaluations of health care interventions. Hence, its use can facilitate informed decision making in health care.

4.1 Background

The attention for informal care appears to be increasing, given the inherent and increasingly noticed scarcity of formal health care resources in many Western countries [158]. Informal care is an important part of total care, especially in the context of chronic illness and frailty due to ageing, and is often provided voluntarily by family, friends or acquaintances. Informal care may reduce the pressure on the capacity and budget of formal health care [61-63]. Moreover, it may be preferred by both patient and informal caregiver over formal care [28]. Notwithstanding the fact that providing informal care can be rewarding [28, 30], caring can have considerable negative effects on the health and general well-being of informal caregivers [21, 42, 48-50, 159, 160]. Therefore, the impact of providing informal care on caregivers should be recognised by policy makers when making decisions concerning the structure and provision of health care services. Moreover, information on the impact of informal care is valuable input for policy decisions regarding arrangements facilitating and supporting informal caregiving in health care.

Economic evaluations aim to support optimal allocation of scarce health care resources. Although inclusion of informal care in economic evaluations is highly desirable [74, 75, 77, 78], at present informal care commonly is ignored in economic evaluations. Thus, policymakers remain ignorant of the impact of interventions in health care on informal caregivers and risk making non-optimal decisions. Moreover, in the few instances that informal care is included in economic evaluations, the comparability of results is hampered by differences in measuring and valuing informal care [25, 91, 92]. This is, for example, reflected in different approaches to measure and value caregivers' time input [25, 26, 91, 99], health [50, 53], and well-being [91, 119].

Common approaches to value informal care, such as the willingness to pay method or the proxy good method, typically provide limited information regarding the underlying informal care situation and its potentially diverse impact [25, 29, 34, 100]. Subjective burden measures for informal care focus more on this latter issue. Several generic and disease-specific subjective burden instruments are available describing the negative impacts of caring, such as problems experienced with mental health, physical health, or social and financial aspects [26, 80, 104, 105, 107, 161]. Some instruments aim to capture the positive impacts of caring as well [29, 33, 105, 108, 110].

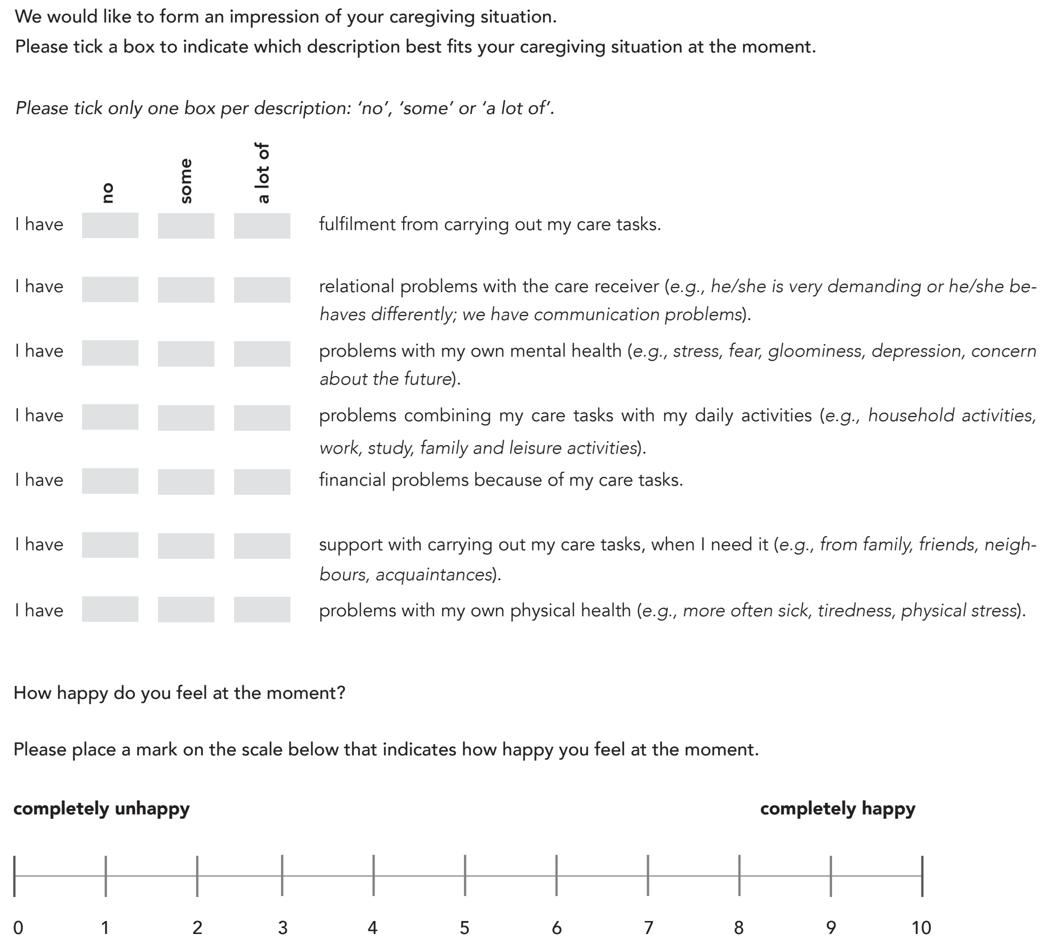
While many of these subjective burden instruments provide a detailed description of caregiving burden, they do not value the impact of caregiving in economic terms, making them unsuitable for economic evaluations. At this time, only two instruments combine an economic valuation of informal care with the informational density of burden instruments: The Carer Experience Scale (CES; [108, 113]) and Care-related Quality of Life instrument (CarerQol; [110]). Both instruments describe the care situation in terms of the negative and positive impact of caregiving, and value the overall impact of informal care. The CarerQol instrument values this impact in two ways: general well-being and care-related quality of life. The (latter) utility scores for the CarerQol are based on preference information from the general public in the Netherlands [162]. The CES instrument values the impact of caregiving with care-related quality of life scores, based on preference information from caregivers of elderly persons in the UK for the CES [108, 113].

When patient interventions are compared in economic evaluations, the CES or CarerQol can be used as an additional source of information in cost-effectiveness analyses using conventional outcome measures, such as patient Quality Adjusted Life Years (QALYs), or as one of the principal outcome measures in cost-consequence or multi-criteria analyses. Furthermore, cost-effectiveness analyses of interventions or support programmes targeted directly at informal caregivers can apply the CES or CarerQol as main outcome.

The focus in this chapter is on the CarerQol (see Figure 4.1). This instrument was developed in 2006, in a similar way as the EuroQol instrument [122], and it has been applied in several studies since [54, 100, 163-168]. Four validation studies of the CarerQol have been conducted previously. Brouwer et al. (2006) and the study reported in chapter 3 performed tests of the feasibility of the CarerQol instrument. Construct validity of the CarerQol instrument was studied in different caregiver samples: two heterogeneous groups of caregivers that were members of regional caregiver support centres in the Netherlands (n=175) in Brouwer et al. (2006) and in chapter 2 (n=230), caregivers of persons permanently living in or receiving day care from a nursing home in the Netherlands in chapter 3 (n=108) and a sample of children with craniofacial malformations living in the US (n=65) [54]. Test-retest reliability of the CarerQol was also investigated in the sample of caregivers of nursing home care patients in chapter 3. These various tests of the psychometric properties of the CarerQol showed favourable results concerning its feasibility in Brouwer et al. (2006) and in chapter 3, construct validity in Brouwer et al. (2006), Payakachat et al. (2011) and in chapters 2 and 3, and test-retest reliability (chapter 3).

The results of these four studies require further confirmation for several reasons. First, the validation studies so far used caregiver samples that were relatively small: the number of respondents ranged between 65 and 230. Secondly, these samples were either overrepresented by relatively strained caregivers in Brouwer et al. (2006) and chapter 2 or by caregivers in a specific informal care situation (e.g., caring for young disabled children [54] or institutionalized elderly (chapter 3)). Thirdly, the range of tests used for construct validation was limited. Most tests concerned whole sample analyses and did not investigate possible heterogeneity among caregivers (because of study sample size). Moreover, few other subjective burden instruments were available from these studies to compare the CarerQol with (e.g., chapter 3, [54]). The study presented in this chapter aimed to overcome most of these shortcomings by using a much larger, heterogeneous sample of informal caregivers (n=1,244), representing a broad range of informal care situations and levels of caregiving burden. This sample size also allows for the construct validation of the CarerQol to be tested in a more elaborate way, which is important given the lack of a gold standard for the impact of caregiving. These tests will be conducted by comparing the performance of the CarerQol with a number of other subjective burden instruments and a range of subgroup analyses comparing between groups of caregivers characterised by differences in caregiving strain, health and socio-economic characteristics.

This study was specifically designed to validate the CarerQol and to demonstrate its ability to assess the overall impact of caregiving in diverse types of caregiving situations. The availability of a validated instrument to measure and value the impact of caregiving will support its application in informal care research. Moreover, it facilitates the inclusion of informal care impacts in economic evaluations of diverse patients and caregiver interventions, and better evidence-based decision making in health care.



We would like to form an impression of your caregiving situation.
Please tick a box to indicate which description best fits your caregiving situation at the moment.

Please tick only one box per description: 'no', 'some' or 'a lot of'.

	no	some	a lot of	
I have	☐	☐	☐	fulfilment from carrying out my care tasks.
I have	☐	☐	☐	relational problems with the care receiver (*e.g., he/she is very demanding or he/she behaves differently; we have communication problems*).
I have	☐	☐	☐	problems with my own mental health (*e.g., stress, fear, gloominess, depression, concern about the future*).
I have	☐	☐	☐	problems combining my care tasks with my daily activities (*e.g., household activities, work, study, family and leisure activities*).
I have	☐	☐	☐	financial problems because of my care tasks.
I have	☐	☐	☐	support with carrying out my care tasks, when I need it (*e.g., from family, friends, neighbours, acquaintances*).
I have	☐	☐	☐	problems with my own physical health (*e.g., more often sick, tiredness, physical stress*).

How happy do you feel at the moment?

Please place a mark on the scale below that indicates how happy you feel at the moment.

Figure 4.1 The CarerQol instrument

4.2 Methods

4.2.1 Data

Data was gathered using an online questionnaire in October 2010. A sample that was representative of the adult Dutch population in terms of age and gender was recruited from a large online panel. From this sample informal caregivers were selected. This was done by asking whether respondents (i) provided care or support, on a voluntarily basis, to a family member, friend or acquaintance who needed help due to physical or mental health problems or problems due to aging, and (ii) for how long they have been lending this care. These selection questions ensured that data would only be gathered among respondents who had been lending informal care for more than two weeks. The questionnaire on informal care was completed by 1,288 respondents of which 44 were dropped from the final sample for analysis. Main reasons were an unrealistically short completion time (i.e., respondents rushing through the questionnaire) or the fact that the answers indicated the respondent was not an informal caregiver after all. The latter was typically the case when the answers indicated a respondent worked for a voluntary organization or provided zero hours of care per week. This left 1,244 questionnaires in the final sample.

4.2.2 Questionnaire

The questionnaire was based on the iMTA Valuation of Informal Care Questionnaire (iVICQ) [162] and included questions on the impact of caregiving as well as characteristics of the caregiver, care recipient and the care situation. The impact of caregiving was measured with the CarerQol instrument, ASsessment of the Informal care situation Scale (ASIS), the Self-Rated Burden scale (SRB), the Process Utility measure (PU), the Caregiver Strain Index (CSI) and Perseverance time (Pt). The CarerQol measures well-being (CarerQol-VAS) and subjective burden (CarerQol-7D). Well-being is measured in terms of happiness using a visual analogue scale (VAS) with endpoints 'completely unhappy' (0) and 'completely happy' (10) (CarerQol-VAS) [110]. Subjective burden is measured on seven dimensions (CarerQol-7D): fulfilment (positive dimension), relational problems (negative dimension), mental health (negative), daily activities problems (negative), physical health (negative) and support (positive). Respondents describe their caregiving situation by selecting one of three possible responses on each dimension: (i) no, (ii) some, and (iii) a lot. The combination of dimensions and answering categories discerns a total of 2187 (= 3^7) caregiving situations. Tariffs are available to compute a weighted sum score for the CarerQol-7D, which represents informal care situation utilities ranging from 0 (worst informal caregiving situation) to 100 (best informal caregiving situation) (chapter 7, [162]). Like common health-related quality of life measures for patients [130, 131] the tariffs for the CarerQol-7D were based on preferences of the general public for different care situations as described with the seven dimensions (and three levels) of the instrument. The ASIS measures the desirability of the caregiving situation with a horizontal VAS ranging from (0) the 'worst imaginable caregiving situation' to (10) the 'best imaginable caregiving situation' (chapter 3). The SRB measures the subjective burden of informal care with a horizontal VAS ranging from (0) 'not straining at all' to (10) 'much too straining' [29, 105]. PU measures the value attached to the process of caregiving by comparing caregivers' current well-being with their well-being in a hypothetical situation that all caregiving tasks would be taken over by someone selected by the caregiver and care recipient, free of costs [28]. The CSI [107] measures the strain of caregiving by asking caregivers' experiences in 13 common problem areas, leading to a non-weighted sum score ranging from 0 to 13. Higher scores indicate higher burden, and caregivers are considered to experience substantial strain when their score is 7 or higher [107]. In addition, the CSI was combined with the five positive aspects of caregiving that caregivers may experience, forming the CSI+ as proposed by Al-Janabi et al. (2010). Finally, Pt indirectly measures the burden of caregiving by asking caregivers how long they expect to be able to continue performing their current informal care tasks if the care situation remains stable, with pre-defined answer categories ranging from less than two weeks to more than two years [169].

Informal caregiver characteristics collected were age, gender, highest attained educational level, performing paid work, household income, health (using EQ-5D [122]) and having a partner. Care recipient characteristics included age, gender, health (using EQ-5D [122]), level of independence (using KATZ scale [153, 170]), need for 24/7 surveillance and type of health problem (defined as a temporary or chronic condition). The care situation was described by the relationship and co-residence of care recipient and caregiver, the number of years, days per week and hours per day caregiving, use of home care, day care, or other institutional care, being on waiting list for day or nursing care, and support from other informal caregivers.

4.2.3 Statistical analyses

Descriptive statistics in percentages and means were calculated of the characteristics of caregivers, care recipients and informal care situations and of the instruments measuring the impact of informal care.

Three types of construct validation were studied: clinical, convergent and discriminative validity. Clinical validity concerns the extent to which the measure relates to variables, such as important background characteristics [98]. Convergent validity is assessed by considering whether a construct of a measure resembles that of other instruments with the same subject of measurement [98]. Discriminative validity tests can be used to study whether 'extreme' groups of respondents score differently on an instrument [98].

4.2.3.1 Clinical validity

Clinical validity was investigated by studying the association between CarerQol-VAS scores and background characteristics of the caregiver, care recipient and informal care situation with one-way ANOVA tests and Spearman's correlation coefficients. Multivariate associations were analysed with ordinary least squares regression (OLS) of CarerQol-VAS scores and caregiver, care recipient and informal care situation characteristics, correcting for subjective burden (CarerQol-7D). Statistically insignificant variables were excluded from the model. We used a less restrictive p-value of 0.2 for this, to avoid excluding variables that did explain variance in CarerQol-VAS scores [171]. Reference values for categorical variables were those with the highest CarerQol-VAS score. To avoid the problem of too few respondents per category, categories of variables were merged if one of the categories represented 10% or less of the data. Missing values for duration of caregiving (missing value in 7 cases) and income (missing value in 342 cases) were supplemented in the multivariate analysis using the multiple imputation by chained equations (MICE) command in Stata® [172, 173] (StataCorp LP).

4.2.3.2 Convergent validity

Convergent validity of the CarerQol was studied by (i) the association between the two parts of this instrument, and by (ii) the association between the CarerQol and other instruments measuring the impact of caregiving included in the questionnaire: ASIS, SRB, PU, CSI and Pt. Spearman's correlation coefficients were used to study bivariate associations of CarerQol-VAS and CarerQol-7D. Multivariate associations were studied using OLS. Subgroup analyses of the multivariate associations of CarerQol-VAS and CarerQol-7D were performed based on a low or high score on the ASIS, SRB, PU, CSI or Pt. Background characteristics that appeared important in explaining CarerQol-VAS (in clinical validity) were also used to construct subgroups. Furthermore, associations of CarerQol-VAS and CarerQol-7D dimensions with ASIS, SRB, PU, CSI and Pt were inspected using Spearman's correlation coefficients. Correlations <0.1 were considered as trivial; 0.1–0.3 as small; 0.3–0.5 as moderate; 0.5–0.7 as high; 0.7–0.9 as very high; >0.9 as nearly perfect [174].

4.2.3.2 Discriminative validity

Discriminative validity of the CarerQol was investigated by contrasting extreme subgroups [98]. Specifically, differences in mean scores on the instruments measuring the impact of caregiving among respondents scoring a 'no' or 'a lot' on CarerQol-7D dimensions were studied with Student's t-tests. All statistical analyses were performed with Stata® version 11.0 (StataCorp LP).

4.2.4 Ethics

No ethics approval was required for this study. We collected data from an online panel. People subscribing to this panel are informed about privacy and data use issues. After deciding to subscribe, people regularly receive invitations to participate in all sorts of online surveys and they are free to accept any invitation they receive. In the case of this study, people received information about the purpose of the study and the organization conducting it (our institute), the type of questions and the estimated completion time. People accepting the invitation were directed to the online questionnaire. After starting completion, they were free to terminate their participation at any point. People submitting their data at the end of the questionnaire were assumed to approve of the content of the questionnaire and their response, and to give consent for use of their response for the purpose of this study, as stated in the invitation. People received a small incentive for completing a questionnaire: after submitting their data, they were offered the opportunity to donate a small sum, depending on the length of the questionnaire, to a charity of their choice. The data received from the survey sampling organization was anonymous.

4.3. Results

4.3.1 Study sample

Table 4.1 presents informal caregiver, care recipient and care situation characteristics. The mean age of caregivers was 47 years. A slight majority of them was female. Somewhat more than fifty per cent had a paid job. Care recipients were on average 63 years old and two thirds of them were female. Their average EQ-5D score was 0.5. Most caregivers lent care to their parents (in-law). On average, caregivers had been providing care for 5 years and spent 18 hours per week on care.

4.3.2 Instruments

The mean CarerQol-7D score was 79.1. The majority of caregivers derived a lot of fulfilment from caregiving. Problems most often encountered were physical health problems and problems with daily activities. Around one third had relational, mental health or financial problems. Most of the caregivers experienced only mild problems. Just over one fourth did not receive support with caregiving when needed. The mean CarerQol-VAS score was 7.1.

The mean ASIS score was 6.7 and the mean SRB score was 4.1. Overall, PU was positive with a mean of 1.6 (implying that the happiness of these caregivers would drop with 1.6 point when handing over all care tasks) and two thirds of caregivers indicated to have a positive PU score. The mean CSI score was 4.8 and 29 per cent experienced 'substantial strain'. On average, caregivers expected to be able to persevere with their care task for two years or more (Table 4.2).

Table 4.1 Sample characteristics and association with CarerQol-VAS; n=1,244

		% or mean	CarerQol-VAS mean	p-value [a]	Spearman's rho	std. coef [b]
Caregiver						
Age	<47.1 years	47.1	7.2	0.20	-0.04	-0.06
	≥47.1 years		7.1			
Gender	female	58.3	7.0	0.02	-	-
	male	41.7	7.3			
Educational level	low	14.6	7.2	0.83	-	-
	middle	55.9	7.1			
	high	29.6	7.1			
Paid work	full-time	26.7	7.4	0.00	-	ref.
	part-time	27.0	7.2			-0.06
	no	46.3	6.9			-0.07*
Income (1-15) [c]					0.11***	
Income [d]	low	31.7	7.0	0.04	-	-0.10*
	middle	24.0	7.1			-0.07
	high	16.9	7.4			ref.
	missing	27.5	7.1			
EQ-5D score	<0.8	0.84	6.4	0.00	0.35***	0.08*
	≥0.8		7.5			
Having a partner	yes	68.9	7.2	0.09	-	-
	no	31.1	7.0			
Care recipient						
Age	<63.6 years	63.6	7.0	0.01	0.06*	0.06
	≥63.6 years		7.2			
Gender	female	66.2	7.2	0.04	-	-
	male	33.8	7.0			
EQ-5D score	<0.5	0.5	6.8	0.00	0.21***	0.09**
	≥0.5		7.4			
Level of independence (1-6)	<4.3	4.3	7.2	0.03	0.06*	-
	≥4.3		7.0			
Surveillance needed 24/7	yes	11.9	6.9	0.10	-	-
	no	88.1	7.2			
Type of health problem	temporary condition	10.3	7.5	0.01	-	-
	chronic condition	89.7	7.1			
Care situation						
Relationship with care recipient	partner	15.3	7.0	0.02	-	0.02
	parent (-in-law)	41.6	7.0			-0.09*
	other family	24.4	7.2			-0.01
	friend / acquaintance	18.8	7.4			ref.
Care recipient shares household	yes	31.0	7.0	0.02		-
	no	69.1	7.2			
Total years care	<5.1 years	5.1	7.2	0.05 [e]	-0.05	-
	≥5.1 years		7.0			
Days p/wk	<4.1 days	4.1	7.2	0.12	-0.04	-
	≥4.1days		7.1			
Hours p/wk	<18.4 hours	18.4	7.2	0.10	-0.06*	-
	≥18.4 hours		7.0			
Professional care [e]	yes	49.6	7.0	0.00	-	-
	no	50.3	7.2			
Day care [e]	yes	8.7	6.9	0.22	-	-
	no	91.3	7.1			
Other informal caregivers	yes	31.4	7.2	0.09	-	-
	no	68.7	7.1			
Institutionalization	yes	24.8	7.1	0.83	-	-
	no	75.2	7.1			
Waiting list [e]	yes	11.9	7.1	0.89	-	-
	no	88.1	7.1			

Note: * p<0.05, ** p<0.01, *** p <0.001; [a] oneway anova test; [b] from multivariate regression analysis corrected for CarerQol-7D; [c] continuous variable, missings not included; [d] categorical variable; [e] caregivers of non-institutionalized care recipients only, n=1062

Table 4.2 Descriptive statistics of CarerQol, ASsessment of Informal care Situation (ASIS), Self-Rated Burden (SRB), Process Utility (PU), Caregiver Strain Index (CSI) and Perseverance time (Pt), n=1,244

		mean (SD)	**%**
CarerQol-7D (0-100)		79.1 (18.6)	
Fulfilment	no		6.7
	some		30.9
	a lot		62.5
Relational problems	no		64.7
	some		28.9
	a lot		6.4
Mental health problems	no		58.2
	some		31.2
	a lot		10.5
Problems combining daily activities	no		50.3
	some		39.1
	a lot		10.6
Financial problems	no		67.9
	some		23.9
	a lot		8.3
Support	no		27.0
	some		47.2
	a lot		25.8
Physical health problems	no		49.4
	some		36.4
	a lot		14.2
CarerQol-VAS (0-10)		7.1 (1.6)	
ASIS (0-10)		6.7 (1.9)	
SRB (0-10)		4.1 (2.5)	
PU	score	1.6 (2.8)	
	positive		66.5
	neutral		8.8
	negative		24.8
CSI	score	4.8 (3.2)	
	substantial burden (>6)		29.1
Pt	in months	23.4 (0.3)	
	<1 week		2.7
	<1 month		3.1
	<6 months		7.5
	>6 months, but <1 year		8.4
	>1 year, but <2 years		9.8
	>2 years		68.6

4.3.3 Clinical validity CarerQol-VAS

Bivariate analyses of CarerQol-VAS scores and background characteristics (Table 4.1) showed that CarerQol-VAS was higher among male caregivers, caregivers with a paid job, in particular a full-time position, caregivers with high income, and caregivers in relatively good health. CarerQol-VAS seemed higher among caregivers lending care to older, healthier or female care recipients. Care situation characteristics that had a bivariate association with CarerQol-VAS were duration and intensity of caregiving, relationship between caregiver and care recipient and sharing a household. Multivariate analysis showed that caregivers' and care recipients' health status and having a full-time job were positively associated, and a low income or giving care to parents (in-law) were negatively associated with CarerQol-VAS.

4.3.4 Convergent validity

4.3.4.1 CarerQol-VAS & CarerQol-7D

Table 4.3 shows that CarerQol-VAS was positively associated with the positive dimensions of the CarerQol-7D fulfilment and support, and negatively with the negative dimensions of CarerQol-7D. Multivariate association of CarerQol-VAS and CarerQol-7D (Table 4.4) confirmed that CarerQol-VAS score were higher among caregivers that experienced fulfilment and received support and when problems were absent. However, relational and financial problems were insignificant in this model.

Table 4.3 Spearman's correlation coefficients of the CarerQol instrument and ASsessment of Informal care Situation (ASIS), Self-Rated Burden (SRB), Process Utility (PU), Caregiver Strain Index (CSI), and Perseverance time (Pt; in months), n=1,244

		CarerQol-VAS	ASIS	SRB	PU	CSI	Pt
CarerQol-VAS		-	0.31	-0.33	0.52	-0.40	0.22
CarerQol-7D	Fulfilment	0.24	0.24	-0.30	0.31	-0.25	0.29
	Relational problems	-0.19	-0.25	0.35	-0.28	0.38	-0.26
	Mental health problems	-0.36	-0.24	0.39	-0.32	0.47	-0.30
	Problems combining daily activities	-0.27	-0.25	0.47	-0.30	0.52	-0.36
	Financial problems	-0.24	-0.24	0.30	-0.22	0.42	-0.26
	Support	0.14	0.13	-0.10	0.09	-0.12	0.02
	Physical health problems	-0.35	-0.22	0.42	-0.27	0.48	-0.25

Note: All Spearman's correlation coefficients are statistically significant at a 99% C.I., except the correlation coefficient of 'support' and PU which is significant at a 95% C.I, and 'support' and Pt which is ns.

Subgroup analyses (Table 4.4) showed that problems with daily activities and support were associated with CarerQol-VAS among relatively burdened caregivers. In addition, problems with daily activities were associated with CarerQol-VAS among caregivers in relatively bad health, and fulfilment among those in relatively good health. Among caregivers of care recipients with a relatively good health status, fulfilment was associated with CarerQol-VAS, and problems with physical health were associated with CarerQol-VAS in the subgroup of care recipients with bad health.

Furthermore, financial problems were associated with CarerQol-VAS among caregivers indicating a long perseverance time, caregivers of their parents and caregivers with low income, while having physical health problems was associated with CarerQol-VAS among high income caregivers.

4.3.4.2 CarerQol & ASIS, SRB, PU, CSI, Pt

Table 4.3 presents the Spearman's correlation coefficients of the CarerQol and the five other instruments. CarerQol-VAS had a positive association with ASIS, PU and Pt, and a negative association with SRB and CSI. CarerQol-7D's positive dimensions had a statistically significant positive association with ASIS, PU and Pt, and a negative one with SRB and CSI. CarerQol-7D support and Pt were not significantly associated. The negative CarerQol-7D dimensions were all negatively associated with ASIS, PU and Pt, and positively with SRB and CSI.

Concerning convergent validity of single CarerQol-7D dimensions (results not presented in a table), the CarerQol-7D item fulfilment had a positive association with CSI 'happy to care' (correlation coefficient (cc) 0.27) and 'care is important' (cc 0.23). CarerQol-7D dimension relational problems was associated with 'recipient appreciates care', 'emotional adjustments', 'behaviour upsetting' and 'recipient change upsetting' (absolute range cc 0.25-0.43). CarerQol-7D dimension mental health was associated with 'emotional adjustments', 'behaviour upsetting', 'recipient change upsetting', 'sleep disturbed', 'inconvenient' and 'feel completely overwhelmed' (absolute range cc 0.18-0.40). CarerQol-7D dimension daily activities was associated with 'confining', 'enough time for self',' family adjustments', 'changes in personal plans' and 'work adjustments' (absolute range cc 0.29-0.47). CarerQol-7D's financial problems was positively associated with CSI 'work adjustments' (cc 0.30) and 'financial strain' (cc 0.57). CarerQol-7D physical health problems was associated with 'inconvenient', 'feel completely overwhelmed', 'physical strain', 'handle the care fine' and 'sleep disturbed' (absolute range cc 0.24-0.40).

4.3.5 Discriminative validity

Table 4.5 shows the mean values of CarerQol-VAS, ASIS, SRB, PU, CSI and Pt per extreme level of CarerQol-7D. Respondents with a lot of fulfilment or support, or no problems on the negative dimensions, had statistically significant higher mean CarerQol-VAS, ASIS, and PU scores, and lower SRB and CSI scores than respondents scoring the other extreme level. The same result applied to Pt, however there was no statistically significant difference in perseverance time among caregivers receiving no or a lot of support. All respondents with a lot of problems on negative CarerQol-7D dimensions experienced 'substantial strain' on the CSI. Caregivers with no fulfilment or no support had a mean CSI value lower than the CSI cut-off point for substantial strain.

Table 4.4 Multivariate regression analysis of CarerQol-VAS, standardized coefficients

			subgroup analyses																	
CarerQol-7D		whole sample	ASIS		SRB		PU		CSI		Pt		EQ-5D score caregiver		EQ-5D score care recipient		income			
			low	high	low	high	low	high	low	high	low	high	low	high	low	high	low	high		
			(n=484)	(n=421)	(n=465)	(n=413)	(n=417)	(n=409)	(n=508)	(n=362)	(n=391)	(n=853)	(n=457)	(n=787)	(n=430)	(n=439)	(n=394)	(n=210)		
Fulfilment	no	-0.12***	0.02	-0.12*	-0.20***	0.11*	-0.13**	0.01	-0.19***	-0.13**	0.06	-0.18***	-0.04	-0.20***	-0.06	-0.28***	-0.10*	-0.17*		
	some	^{a}	-0.12**	^{a}	^{a}	-0.10*	^{a}	^{a}	^{a}	^{a}	-0.08	^{a}	^{a}	^{a}	^{a}	^{a}	^{a}	^{a}		
Relational	some	-0.01	-0.06	0.02	-0.01	-0.06	0.04	-0.02	0.04	-0.05	-0.01	-0.02	-0.02	-0.02	-0.01	0.01	0.03	0.08		
problems	a lot	^{b}	0.00	^{b}	^{b}	0.00	0.04	^{b}	^{b}	-0.03	-0.03	^{b}	^{b}	^{b}	^{b}	^{b}	^{b}	^{b}		
Mental health	some	-0.14***	-0.13**	-0.23***	-0.18**	-0.09	-0.18**	-0.08	-0.15**	-0.05	-0.10	-0.21***	-0.19***	-0.06	-0.13*	-0.22***	-0.21***	-0.06		
problems	a lot	-0.21***	-0.28***	^{b}	^{b}	-0.36***	-0.38***	^{b}	^{b}	-0.22**	-0.21**	^{b}	-0.36***	^{b}	-0.32***	^{b}	-0.27***	-0.24**		
Problems with	some	-0.04	-0.01	-0.06	-0.03	-0.02	0.02	-0.05	-0.01	-0.13**	-0.06	-0.02	-0.01	-0.06	-0.02	-0.09 ^{c}	-0.07	-0.09		
daily activities	a lot	-0.08**	-0.17**	-0.05	^{b}	-0.08	-0.03	^{b}	^{b}	-0.27***	-0.12	^{b}	-0.15*	^{b}	0.00	^{b}	^{b}	-0.03		
Financial	some	-0.03	0.03	-0.06	-0.01	0.01	-0.02	-0.05	-0.09	0.00	0.01	-0.07*	-0.01	-0.08*	-0.03	-0.03	-0.12*	0.07		
problems	a lot	^{b}	0.03	^{b}	^{b}	0.05	-0.00	^{b}	^{b}	0.00	0.01	^{b}	-0.01	^{b}	0.01	^{b}	-0.07	^{b}		
Support	no	-0.11***	-0.15**	-0.05	-0.06	-0.14*	-0.15**	-0.05	-0.05	-0.15*	-0.20**	-0.06	-0.12*	-0.10*	-0.12*	-0.10*	-0.09	-0.03		
	some	-0.07**	-0.06	-0.10	-0.04	-0.07	-0.11	-0.03	-0.03	-0.09	-0.13*	-0.03	-0.10*	-0.05	-0.05	-0.07	-0.09	-0.06		
Physical health	some	-0.12***	-0.13*	-0.11 c	-0.14**	-0.14*	-0.07	-0.17**	-0.14**	-0.17*	-0.10	-0.14**	-0.05	-0.13**	-0.11	-0.08	-0.09	-0.18*		
problems	a lot	-0.13***	-0.15**	^{b}	^{b}	-0.22***	-0.13*	^{b}	^{b}	-0.18*	-0.19**	^{b}	-0.04	^{b}	-0.13*	^{b}	-0.04	-0.13		
Constant		8.13	7.96	8.34	8.27	8.04	7.60	8.33	8.15	8.34	8.04	8.11	7.78	8.14	7.84	8.41	7.39	8.31		
Adjusted R²		0.21	0.25	0.16	0.14	0.25	0.23	0.07	0.13	0.21	0.17	0.18	0.17	0.14	0.16	0.25	0.23	0.24		

Table 4.5 Mean values of CarerQol-VAS, ASsessment of Informal care Situation (ASIS), Self-Rated Burden (SRB), Process Utility (PU), Caregiver Strain Index (CSI) and Perseverance time (Pt; in months) per 'extreme level' of CarerQol-7D; n=1,244

CarerQol-7D		CarerQol-VAS	ASIS	SRB	PU	CSI	Pt
Fulfilment	no (n=83)	6.9	5.9	5.9	0.6	6.4	11.7
	a lot (n=777)	7.4	7.0	3.6	2.3	4.2	25.6
Relational problems	no (n=805)	7.4	7.0	3.5	2.2	3.9	25.4
	a lot (n=80)	6.6	5.6	6.0	0.0	7.4	18.4
Mental health problems	no (n=724)	7.6	7.1	3.3	2.3	3.5	25.8
	a lot (n=131)	5.7	5.7	6.1	-0.4	7.7	16.5
Problems combining daily activities	no (n=626)	7.5	7.2	3.0	2.4	3.2	26.8
	a lot (n=132)	5.9	5.8	6.4	-0.2	8.2	14.5
Financial problems	no (n=844)	7.4	7.0	3.6	2.0	3.8	25.2
	a lot (n=103)	6.5	5.7	5.7	0.7	7.7	16.5
Support	no (n=336)	6.9	6.5	4.3	1.5	5.2	23.3
	a lot (n=321)	7.5	7.2	3.6	2.1	4.1	23.7
Physical health problems	no (n=614)	7.7	7.1	3.1	2.3	3.3	25.6
	a lot (n=177)	6.1	5.8	6.0	0.2	7.2	17.3

Note: All differences in means are statistically significant at a 99% C.I., except Support & PU: significant at C.I. 95%, Support & Pt is ns.

4.4 Discussion

This study investigated whether the CarerQol validly assessed the overall impact of caregiving in a large, heterogeneous sample of caregivers from the Netherlands. Results of clinical, convergent and discriminative validity tests confirmed the favourable results from previous studies. Both the subjective burden (CarerQol-7D) and the well-being (CarerQol-VAS) component of the CarerQol were significantly associated in the expected direction with other measures of the impact of caring. Additionally, well-being was related to important caregiver, care recipient and care situation characteristics, in expected directions. Hence, this study adds to previous evidence suggesting that the CarerQol may be a useful measure of the impact of caregiving in research in a wide variety of informal care contexts. Moreover, it facilitates inclusion of informal care in economic evaluations of health care interventions.

The CarerQol instrument measures care-related quality of life of caregivers. This concept is broader than the generally used outcome measure in economic evaluations, as for instance health-related quality of life in cost-utility analyses. Therefore, the results of the CarerQol cannot be easily combined with patient outcomes cost-effectiveness or -utility analyses. Nevertheless, as stated in the Introduction, the results of the CarerQol can be presented alongside the results of an economic evaluation and so inform decision makers more completely about the total impact of an intervention on society. The CarerQol can also be used with other types of economic evaluation, such as multi-criteria analyses, which allow considering multiple outcome measures. Finally, the CarerQol can very well serve as the main outcome measure in economic evaluations of programmes for caregivers (e.g., support programmes, respite care).

Before the implications of the results will be discussed in more detail, it is important to note some limitations of this study. First, although a representative sample of the adult Dutch population in terms of age and gender was used, the study sample may be somewhat selective due to the use of an online panel. Internet was considered to be a suitable medium to gather data, because more than 90 per cent of the Dutch population uses internet. In addition, while elderly may be a typical group expected to be underrepresented in internet surveys, it needs noting that in recent years elderly became more active on the internet with six out of ten elderly of 65 to 75 years currently being internet users [175]. Furthermore, selection bias could have occurred in using an online panel to select caregivers. We did not have a priori reasons to suspect that caregivers in general would be less likely to subscribe to online panels. This may be the case for caregivers experiencing high levels of strain, but this group is generally difficult to approach and less likely to participate in any type of survey. Hence, the subgroup of caregivers experiencing severe strain may be underrepresented in the study sample, but not more or less than in other studies among caregivers. On the other hand, in previous validation studies (chapters 2, 3, [54, 110]) caregivers were recruited via organisations providing information and support services for caregivers. It is likely that people who come to such organisations to ask for assistance see themselves as caregivers and concern a selection of caregivers experiencing relatively higher strain in their caregiving situation. Through the online panel and the selection questions used in this study it is likely that persons lending care were reached who would normally not define themselves as caregivers, for instance because their burden is low, and therefore would not be represented in these previous studies. All in all, the study sample is expected to be more representative of the caregiver population, in particular at the low burden end.

Secondly, in the multivariate models, the multiple imputation method (MI) was used to handle missing values. An assumption of this method is that these values are, at least, missing at random [172, 176]. While income is a classic example of missing not at random [177], MI is nevertheless considered as the best alternative to other strategies, such as excluding respondents with missing values from the model, or mean imputation. In addition, income correlated with other background characteristics in the data, such as gender of the caregiver, which gives some support to the chosen imputation method.

Thirdly, validation is an on-going process, and therefore, testing psychometric properties among caregivers in other settings, such as caregivers in other countries than the Netherlands, as for instance recently in the US [54], remains desirable. In addition, other psychometric properties of the CarerQol could be further tested, such as reliability (chapter 3) and sensitivity to change.

The overall well-being in this sample was relatively high compared to caregiver samples of previous validation studies. Possibly this is due to the recruitment procedure in this study that may have attracted significantly more caregivers in low burden care situations, as discussed before. Higher well-being scores were found among caregivers with positive care experiences in terms of fulfilment from caregiving and assistance from others in lending care, which many caregivers reported to have. Furthermore, as previous CarerQol validation and informal care studies underlined, well-being of caregivers was positively related to health of both the caregiver and care recipient (chapters 2, 3, [53, 103, 110, 178]). Moreover, as other studies on informal care also suggest (e.g., [53, 103]), well-being of caregivers was positively influenced by more general aspects of life, not necessarily (directly) related to caregiving [179], such as having a full-time paid work position or a high income. Besides these positive influences, caregivers also experienced negative consequences of lending care. Important to note here, is that this study used cross-sectional data and hence it was not possible to determine the causality of the established associations. Caregivers often faced problems with their health and also reported difficulties combining care with other daily activities. Having these problems, negatively affected their well-being. Further, especially those caring for their parents (in-law) reported lower happiness scores.

Our study clearly indicates that the diverse problems associated with informal care are not equally important for all caregivers. For example, financial problems were negatively associated with happiness of caregivers with a relatively low income particularly. In addition, mental health problems and problems with daily activities were associated with well-being among caregivers with a relatively low health status, while physical health problems and financial problems were among relatively healthy caregivers. Differences were also observed for receiving support with care tasks. The positive influence of support was especially prevalent among highly burdened caregivers. Furthermore, a note should be made on the CarerQol-7D dimension relational problems, which showed overall satisfactory convergent and discriminative validity results, but did not impact well-being when considered in combination with other burden dimensions. This seems to contradict some previous validity results (chapter 3, [110]). However, additional tests in subgroups, more closely resembling the samples used in earlier studies, confirmed that relational problems did affect well-being among caregivers of recipients with a persistent care need often due to chronic or age-related health problems. This diversity in importance of burden dimensions among subgroups indicates that although some aspects of caregiving burden may not seem relevant in some group of caregivers, they may matter to caregivers in another context, as was described above for the dimension relational problems. Additionally, given that not all problems are equally important for caregivers, it is recommendable that support programmes target the problems that are relevant to specific (groups of) caregivers, such as relieving financial problems of caregivers on low income.

Convergent and discriminative validity tests using the ASIS, SRB, PU, CSI and Pt instruments, which aim to measure a similar construct as the CarerQol, showed that the CarerQol instrument performs well. That is, when the ASIS, SRB, PU, CSI and Pt reported higher caregiving burden, the CarerQol also indicated a higher negative impact of caregiving. All these associations between the CarerQol and the overall scores of the ASIS, SRB, PU, CSI and Pt were as expected and the strength of correlations was small to high. The support dimension of the CarerQol-7D showed the weakest correlation with these other measures.

The associations between single CarerQol-7D dimensions and similar individual items of the CSI confirmed the hypothesized relationships as well. The only hypothesis that was rejected was that of the CarerQol-7D dimension daily activities and the CSI item ‘other demands on time’. This counterintuitive result may be explained by different content of both items, because the CSI item merely registers whether caregivers perform other activities, while the CarerQol-7D focuses on difficulties with combining these with caregiving.

Discriminative validity tests showed that the CarerQol-7D discriminated well between caregivers with low or high burden. These results were less stable for the dimension support. Although in general support is an important issue for caregivers [108], some of the validity results were less satisfactory for the support dimension of the CarerQol. This could be explained by the fact that receiving support could have both a positive and a negative effect on caregiving strain. For example, it has been shown that sharing tasks with other informal caregivers tends to decrease burden, but that cooperation with others may also increase burden in case of disagreements between caregivers [160]. Given that support from family or friends with caregiving can both relieve and intensify strain among caregivers, the overall effect of support can level out when studying a large, diverse group of caregivers. It would be interesting to investigate this in more detail in future studies using the CarerQol, for instance by adding a few supplementary questions about the amount, type and perceived quality of support.

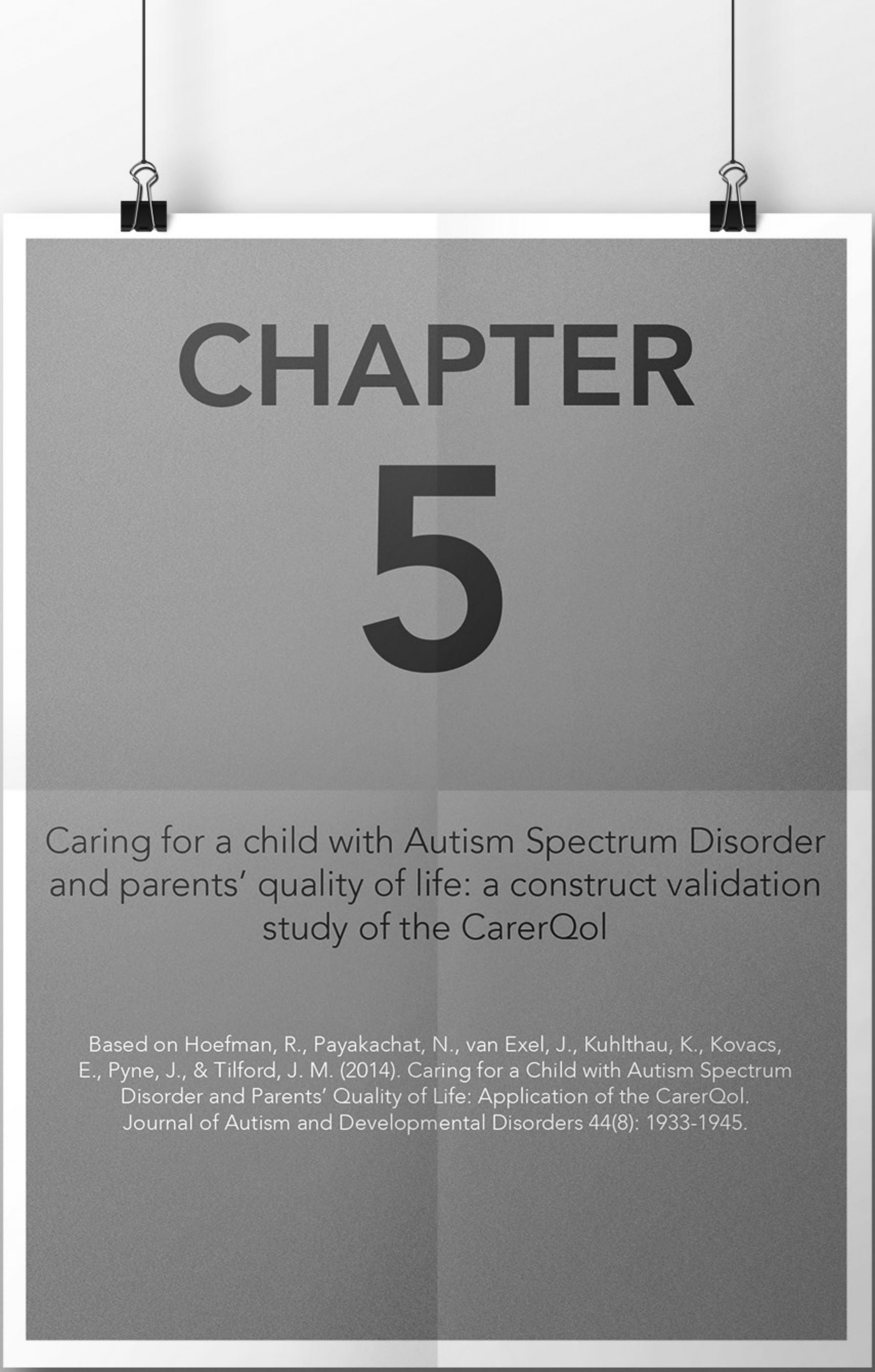
4.5 Conclusions

This study largely confirmed previous findings on construct validation of the CarerQol and added new, strong arguments that this instrument is a valid measure of the overall impact of informal care. Therefore, the CarerQol can be applied in both informal care research and in economic evaluations of diverse patients or caregiver interventions to reveal the important, but often hidden, impact of informal caregiving for well-informed health care policy.

Appendix chapter 4

Table A4.1 Multivariate associations of CarerQol-VAS with CarerQol-7D and background characteristics of caregiver, care recipient and care situation (n=1,244)

		std. coef.	p-value
CarerQol-7D			
Fulfilment		0.08	0.00
Relational problems		0.00	0.87
Mental health problems		-0.21	0.00
Problems with daily activities		-0.10	0.00
Financial problems		-0.02	0.51
Support		0.07	0.01
Physical health problems		-0.09	0.01
Age caregiver		-0.06	0.05
Paid work (ref. fulltime)	part-time	-0.06	0.07
	no	-0.07	0.04
Income (ref. high)	low	-0.10	0.03
	middle	-0.07	0.09
EQ-5D score caregiver		0.08	0.01
Age care recipient		0.06	0.07
EQ-5D score care recipient		0.09	0.00
Relationship (ref. friend /acq.)	partner	0.02	0.52
	parent(-in-law)	-0.09	0.01
	other family	-0.01	0.81
Constant		6.98	
Adjusted R^2		0.23	



CHAPTER

5

Caring for a child with Autism Spectrum Disorder and parents' quality of life: a construct validation study of the CarerQol

Based on Hoefman, R., Payakachat, N., van Exel, J., Kuhlthau, K., Kovacs, E., Pyne, J., & Tilford, J. M. (2014). Caring for a Child with Autism Spectrum Disorder and Parents' Quality of Life: Application of the CarerQol. Journal of Autism and Developmental Disorders 44(8): 1933-1945.

Abstract

This study describes the impact of caregiving on parents of children with autism spectrum disorders (ASDs). Secondly, construct validation of the Care-related Quality of Life instrument (CarerQol) measuring impact of caregiving was investigated. Primary caregivers of children with ASDs were included. Many parents experienced considerable problems combining daily activities with care, had financial problems or suffered from depressive mood. Validity tests showed that a higher impact of caring on the CarerQol was positively associated with higher subjective burden and lower family quality of life. Most of the associations between CarerQol scores and background characteristics confirmed previous research. The CarerQol validly measures the impact of caregiving for children with ASDs on caregivers in the study sample. The CarerQol may therefore be useful for including parent outcomes in research on ASD.

5.1 Introduction

Caring for children with autism spectrum disorders (ASDs) is challenging and affects family life. ASDs are neurological complex conditions impairing social interaction and communication, such as difficulties to respond to social interactions or deficits in understanding nonverbal communication. Moreover, persons diagnosed with an ASD have restricted behavioural functions, such as stereotype behaviour or inability to adjust to new situations [180]. Children with ASDs often exhibit more than one of these core ASD symptoms and many also suffer from associated symptoms, such as severe tantrums or sleep problems [181, 182]. Moreover, health in general of these children is lower than that of typically developing children [183-185]. Meeting the high care demands of affected children requires much time, effort and patience. This often results in psychological distress, depression, anxiety and other mental or physical health problems among their parents [186-189]. Moreover, many parents face financial problems, given high out-of-pocket health care expenses, underemployment or employment loss [186-188, 190]. Not surprisingly, parents of children with ASDs often feel strained by caregiving [189, 191].

Given these prolonged and multidimensional care needs of children with ASDs, it is important to accurately measure the impact of caregiving on the lives of the parents of these children. At present, evaluations of ASD treatments are often limited to measurement of effects in children. However, interventions for children with ASDs often require parents' involvement and also aim to increase parents' caregiving skills, self-efficacy, knowledge of the disorder, and aim to reduce family stress [192]. Furthermore, improved well-being of parents could positively influence the effect these interventions have on children with ASDs [193, 194]. Hence, for fully understanding the effectiveness of ASD interventions it is essential that family outcomes of interventions are also included in evaluation studies [195]. Insights from such studies will help develop interventions focusing on the needs of children with ASDs and their family, and facilitate consideration of those in both policy and funding decisions in health care.

Studies in the field of ASD interventions covering parent outcomes often measure parents' health, in terms of for example, stress, anxiety and mental health [195]. A drawback of this approach is the focus on a single aspect of a caregivers' quality of life, i.e., health-related quality of life. Other important domains of quality of life that can be affected by caring, like social or financial problems, the parent-child relationship or family functioning in general [195] are not considered. In contrast to health-related quality of life, general quality of life considers all domains of quality of life [26, 53]. While there is debate in the literature on the definition of general quality of life, most definitions consider it as a multi-dimensional concept encompassing subdomains such as physical, material, social and emotional well-being [196]. In theory, general quality of life thus could record all possible effects of caregiving. However, such an outcome might be too broad in the context of informal care because it may be difficult to disentangle the effects of caregiving and other aspects affecting general quality of life [53, 103]. To overcome the problem of measuring effects that are not directly related to caregiving, an outcome that focuses directly on the caregiving situation could be used. Subjective burden instruments, for example, report the level of caregiving strain felt by caregivers. Many such instruments are available, like the often used Caregiver Strain Index [107] and Zarit Burden Interview [197]. Among children with mental, emotional or behavioural problems the Caregiver Strain Questionnaire [189, 191, 198] has also been used. These instruments provide detailed information on problems of caregiving, such as disturbed daily patterns or difficulties communicating with the care recipient [26, 104, 105, 161]. However, although caregivers experience strain of dealing with care problems, they may also derive fulfilment from providing care to a loved one and gain abilities from it, such as developing more positive perspectives on life [28, 30-32]. Hence, some subjective burden instruments incorporate both negative and positive aspects of caregiving, such as the Carer Experience Scale [108] and the Self-rated burden scale (SRB; [105]). A drawback of using subjective burden instruments in evaluations of interventions for children with ASDs is that they mainly report problems on separate dimensions of burden and lack an overall subjective burden score. However, evaluative research requires such an overall score to appropriately report and compare the impact of caring on caregivers between interventions. Preferably, this score reflects differences in importance of care problems, because the extent to which specific burden dimensions have an impact on quality of life differs between caregivers [34]. Simple aggregation of burden dimensions to reflect total subjective burden without correcting for the severity of problems experienced by caregivers could lead to misleading conclusions.

The Care-related Quality of Life instrument (CarerQol, see Figure 5.1) was specifically developed to measure outcomes in caregivers for use in evaluative research, and so to provide the essential information for well-informed policy decisions in health care (chapter 8, [110]). The approach of the CarerQol to measure caregiver outcomes was based on the EuroQol instrument measuring health-related quality of life. The CarerQol measures perceived burden in two positive and five negative dimensions, provides a weighted overall subjective burden score and measures general quality of life of caregivers [110]. The CarerQol can be administrated in different caregiver populations, because the questions do not refer to a specific care situation. Such a generic instrument might be less sensitive to very specific problems experienced by caregivers in a particular context, such as parents caring for a child with an ASD, and hence less useful in a clinical setting, but it enables comparisons of the impact of caring between different populations of patients and caregivers. Therefore, the CarerQol facilitates descriptive research on the impact of providing care on a caregiver's life. Moreover, the CarerQol enables comparative research of interventions for children with ASDs including parent outcomes in health care or other sectors, such as education. Finally, interventions directly aimed at parents of children with ASDs, like support programs for caregivers, can be also evaluated with the CarerQol.
The ability of the CarerQol to measure the impact of caring is supported by several construct validation studies, mostly performed in populations of caregivers for adults (chapters 2 and 3, [110]). To date, one set of validity tests of the CarerQol has been conducted among caregivers for children, in the specific context of children with craniofacial malformations [54]. This study investigated associations between the impact of caregiving and the health of caregivers. Moreover, the SRB was used for construct validation of the CarerQol. Results showed that better scores on the CarerQol instrument were associated with better health and lower SRB scores among caregivers of children with craniofacial malformations [54].

5.1.1 Research objectives

This study addresses two research questions. The first objective of this study is to report the impact on parents of caregiving for a child with an ASD. Given the prolonged and multidimensional nature of the care and attention required by the growing child with an ASD, there is a need to better understand the problems encountered by these parents. The second objective of this study concerns construct validation of the CarerQol instrument in a sample of caregivers of children with ASDs.

In other words, this chapter aims to investigate the ability of the CarerQol to measure the impact of providing care on caregivers of children with ASDs. This study is the first to investigate construct validation of the CarerQol among parents of children with ASDs. This study will also add to the knowledge on measurement of the impact of caregiving among caregivers of children in general, by using more elaborate tests than the prior CarerQol validation study among parents [54]; for example, by studying multivariate associations of associates of the CarerQol instrument and performing subgroup analyses among caregivers.

We would like to form an impression of your caregiving situation.
Please tick a box to indicate which description best fits your caregiving situation at the moment.

Please tick only one box per description: 'no', 'some' or 'a lot of'.

	no	**some**	**a lot of**	
I have	2.8%	39.7%	57.5%	fulfilment from carrying out my care tasks.
I have	55.1%	34.6%	10.3%	relational problems with the care receiver (*e.g., he/she is very demanding or he/she behaves differently; we have communication problems*).
I have	41.9%	45.1%	13.0%	problems with my own mental health (*e.g., stress, fear, gloominess, depression, concern about the future*).
I have	39.3%	47.7%	13.0%	problems combining my care tasks with my daily activities (*e.g., household activities, work, study, family and leisure activities*).
I have	47.7%	38.3%	14.0%	financial problems because of my care tasks.
I have	23.6%	50.9%	25.5%	support with carrying out my care tasks, when I need it (*e.g., from family, friends, neighbours, acquaintances*).
I have	44.4%	42.1%	13.4%	problems with my own physical health (*e.g., more often sick, tiredness, physical stress*).

How happy do you feel at the moment?

Please place a mark on the scale below that indicates how happy you feel at the moment.

completely unhappy **completely happy**

Note: Observed score in study sample (mean (SD)): 7.4 (1.9)
4% of the respondents had one or more missing values on the CarerQol instrument.

Figure 5.1 CarerQol instrument and descriptive statistics, n=221

5.2 Methods

5.2.1 Data collection

Data were collected from primary caregivers of children with ASDs who participated in clinical registries at two Autism Treatment Network (ATN) sites in the US: Developmental Center, Little Rock, Arkansas and Outpatient Psychiatric Clinic at Columbia University Medical Center, New York. Families of affected children aged 4 – 17 years were invited to participate in this study by mail. The registry of the ATN site in Arkansas consisted of 247 families. Of these families, 25 did not want to be contacted for research purposes. Two families participated in a pilot survey and were not included in the final survey. The response rate of the final survey was 52.3% (n=115). In New York, 179 families were invited to participate in the survey and 109 of these families completed both questionnaires (response rate 60.9%). In total, 224 families participated in the survey. These families, on average, had two children living in their home with one child having special health care needs and requiring assistance with daily living needs.

The recruitment letter specified that the primary caregiver of the child with an ASD needed to complete two surveys, one as a proxy reporter for the child's perspective (child health questionnaire), the second from their own perspective as caregiver of a child with an ASD (primary caregiver health questionnaire). Respondents were specifically instructed that the health questions in the child health questionnaire pertained to the health of the child with an ASD and in the primary caregiver health questionnaire to the respondents' own health. A $25 gift certificate was provided to a family that returned the two completed surveys. The study protocol was approved by the institutional review boards at Columbia University/New York State Psychiatric Institute and University Arkansas for Medical Sciences.

5.2.2. Child health questionnaire

The child health questionnaire included information on child age, gender, and whether the child was a first child. Child's health status was assessed with the Health Utility Index (HUI-3; [199]) and Quality of Well-being Scale (QWB-SA; [200]). The HUI-3 score ranges from 0 'dead' to 1 'best possible health' based on eight dimensions (vision, hearing, speech, ambulation, dexterity, emotion, cognition and pain). The QWB-SA has a similar range (0 'dead' to 1 'full health') and consists of two parts: (i) questions on mobility, physical activity, social function and (ii) a list of 27 symptoms and problem complexes.

5.2.3 Primary caregiver health questionnaire

The primary caregiver questionnaire included questions on characteristics of (i) the caregiver, (ii) the informal care situation and (iii) the impact of caregiving. First, characteristics of the caregiver included in the questionnaire were age, gender, marital status, highest attained educational level, performing paid work, household income, and health. Caregiver's health was measured with two generic health utility instruments; the EuroQol descriptive system (EQ-5D; [122]) and a six dimensional health state classification, the SF-6D [201]. The EQ-5D consists of five dimensions of health: mobility, self-care, usual activities, pain/discomfort and anxiety/depression. The SF-6D uses six health dimensions: physical functioning, role limitation, social functioning, pain, energy and mental health. Both instruments result in a total score of health with 0 defined as 'dead' and 1 as 'full health'.

In addition, mental health was measured with the Center for Epidemiologic Studies Depression Scale (CES-D; [202]). The CES-D measures depressive feelings and behaviours during the past week using 20 items and has a total score that ranges from 0 to 60. A cut-off point of 16 on the CES-D was used to indicate high level of depressive symptoms [202]. Secondly, questions about the care situation concerned the number of hours 'on call' for caregiving and the presence of other caregivers for the care of children. Thirdly, the impact of caregiving was measured with the CarerQol, the SRB and the Family Quality of Life Scale (FQLS). The CarerQol is comprised of the CarerQol visual analogue scale (CarerQol-VAS) and its descriptive system (CarerQol-7D; [110], see Figure 5.1). The CarerQol-VAS measures general quality of life in terms of happiness using a horizontal visual analogue scale (VAS) with defined endpoints of (0) 'completely unhappy' and (10) 'completely happy' [110]. The CarerQol-7D measures care-related quality of life of caregivers and contains seven dimensions of caregiving burden. Five of these dimensions report the negative aspects of caring: (i) relational problems with the care recipient, (ii) mental health problems, (iii) problems with daily activities, (iv) financial problems, and (v) physical health problems. The CarerQol-7D also reports two positive experiences from caring: (vi) fulfilment and (vii) support. Each of these seven dimensions has three response categories: (i) no, (ii) some, and (iii) a lot. Answers on the negative dimensions of the CarerQol-7D receive a value of 0 (a lot), 1 (some) and 2 (no). Answers on the positive dimensions receive a value of 0 (no), 1 (some), and 2 (a lot) [162]. A scoring mechanism is available to calculate a care-related quality of life score (chapter 7). These weights, or tariffs, are based on preferences for the positive and negative CarerQol-7D dimensions from the general population in the Netherlands. Applying these tariffs to the CarerQol-7D profiles yields a weighted sum score reflecting a specific part of quality of life directly related to caregiving; care-related quality of life (more practical issues concerning the calculation of the CarerQol-7D score can be found in the iMTA Valuation of Informal Care Questionnaire (iVICQ) [162]).

SRB measures subjective burden of informal care and consists of a single horizontal VAS ranging from (0) 'not straining at all' to (100) 'much too straining' asking respondents how burdensome they feel that caring is at the moment [105]. The FQLS captures quality of life of families with children [203]. It contains 25 items divided into 5 subdomains including family interaction (6 items), parenting (6 items), emotional well-being (4 items), physical/material well-being (5 items), and disability-related support (4 items). A total score is calculated by summing all 25 items (ranging from 25 to 125). Subdomain scores are calculated by summing all items on each subdomain with higher scores representing higher traits. A higher total score refers to higher family quality of life.

5.2.4 Statistical analyses first research objective

To answer the first research question on the impact of caregiving on parents of children with ASDs, descriptive statistics in percentages and means (standard deviations; SD) were calculated for the child, parent, and care situation characteristics. Furthermore, the impact of caregiving measured with the CarerQol instrument, the SRB and the FQLS was also presented in percentages and means (SD).

5.2.5 Statistical analyses second research objective

The second research objective of the study was construct validation of the CarerQol. Here, construct validation was conducted by testing convergent, discriminative and clinical validity of the CarerQol [98]. Convergent validity tests assessed whether the construct of the CarerQol instrument resembled that of other instruments with the same subject of measurement. Discriminative validity tests assessed whether 'extreme groups' of respondents had different scores on the CarerQol instrument [98]. Clinical validity tests investigated the extent to which differences in background characteristics were reflected in the scores of the CarerQol instrument. The statistical tests used for the three types of validity will be discussed in the following sub-paragraphs.

5.2.6 Convergent validity tests CarerQol

In this study, convergent validity was studied by investigating the association between (i) the two parts of the CarerQol instrument (i.e., the CarerQol-7D and CarerQol-VAS), and (ii) between the CarerQol instrument and the other two instruments of the impact of caregiving on parents: the SRB and FQLS. First, the association between the CarerQol-VAS and CarerQol-7D was studied with bivariate and multivariate tests. Spearman's correlation coefficients were used for testing bivariate associations of CarerQol-VAS and CarerQol-7D. Multivariate association of CarerQol-VAS and CarerQol-7D was studied with ordinary least squares regression (OLS). These multivariate associations were also tested among subgroups of caregivers of children with ASDs. These subgroups experienced either a relatively low or high impact of caregiving: (subgroup 1, n=120) below average SRB score, (subgroup 2, n=83) equal to or higher than average SRB score, (subgroup 3, n=75) below average FQLS total score, and (subgroup 4, n=120) equal to or higher than average FQLS total score. The following hypotheses were formulated for convergent validity tests of the two parts of the CarerQol instrument. We expected that CarerQol-7D dimensions fulfilment and support were positively associated with CarerQol-VAS. The five negative CarerQol-7D dimensions were expected to have a negative association with CarerQol-VAS.

Secondly, to study associations between the CarerQol and the SRB and FQLS, Spearman's correlation coefficients were used. For these convergent validity tests the following hypotheses applied. A priori, negative associations of CarerQol-VAS with SRB were hypothesized. Moreover, negative associations were also expected for the positive CarerQol-7D dimensions with SRB. Positive associations were hypothesized for the negative CarerQol-7D dimensions and SRB. Positive associations were expected for CarerQol-VAS and FQLS. Furthermore, in this chapter it was expected that positive CarerQol-7D dimensions would positively correlate with FQLS. Negative CarerQol-7D dimensions were hypothesized to negatively correlate with FQLS. The strength of Spearman's correlation coefficients of the convergent validity tests was assessed by the following guideline: <0.1 trivial; 0.1–0.3 small; 0.3–0.5 moderate; 0.5–0.7 high; 0.7–0.9 very high; >0.9 nearly perfect [174].

5.2.7 Discriminative validity tests CarerQol

We analysed discriminative validity of the CarerQol by testing for differences in outcome scores among 'extreme' groups of caregivers. These groups of caregivers were constructed using their answers on the seven burden dimensions of the CarerQol-7D. We constructed groups that did, or did not, report fulfilment or support. The same was done for the five negative CarerQol-7D dimensions and groups were constructed with or without problems on the relational, mental health, daily activities, financial or physical health problem dimensions. Among these seven pairs of 'extreme' groups, differences in the level of impact of caregiving were tested using three instruments: the CarerQol-VAS, SRB and FQLS. Moreover, it was investigated whether caregivers in negative 'extreme' groups had lower health status than those in the positive 'extreme' groups. The same was done for the health status of the children. Differences in mean scores among the extreme CarerQol-7D groups were analysed with Student's t-tests. The following results were expected a priori for the discriminative validity tests of the CarerQol. First, it was hypothesized that caregivers deriving fulfilment or receiving support overall had higher CarerQol-VAS and FQLS scores and lower SRB scores. Secondly, they and their children were expected to be in better health than those without fulfilment or support. A third hypothesis concerned that caregivers experiencing any of the five CarerQol-7D problem dimensions would have lower CarerQol-VAS, FQLS, child health or own health scores and higher SRB scores than those without these care problems.

5.2.8 Clinical validity tests CarerQol

To investigate clinical validity, associations between CarerQol-VAS and background characteristics of the child, caregiver and care situation were studied. Bivariate associations were studied with One-way ANOVA tests. Multivariate association between CarerQol-VAS and these characteristics was studied with OLS. We tested several model specifications. We started with the base model of CarerQol-VAS explained by the dimensions of the CarerQol-7D. The child, caregiver and care situation characteristics were added to this base model. Stepwise backward OLS regression with a cut-off p-value >0.2 was used to delete insignificant child, caregiver and care situation variables from the model. Categories of background variables were merged if they contained less than 10% of the study sample. The category with the highest CarerQol-VAS score was used as reference category for dummy variables. All statistical analyses were performed with Stata® version 12.1 (StataCorp LP).

5.3 Results

5.3.1 Sample characteristics children and caregivers

Our sample included 224 parents of children with ASDs (n=109 ATN site in New York, n=115 ATN site in Arkansas). The children in the study sample were on average 8.4 years (SD 3.5) old (Table 5.1). The majority of children was male (87%). Children's mean HUI-3 score was 0.66 (SD 0.23). Their mean QWB-SA score was 0.59 (SD 0.16).

Caregivers of children with ASDs predominantly were the biological parents (95%), mostly mothers (89%) (Table 5.1). On average, caregivers were 39.4 years (SD 8.3) old. About sixty percent had either a college, professional or graduate school degree and many had a paid job. Somewhat more than one third of families of the children with ASDs had an income of more than $100,000 per year. On average, caregivers' subjective health on a scale from 0 to 100 was 77.6 (SD 15.8), their EQ-5D score was 0.85 (SD 0.14) and their SF-6D score was 0.74 (SD 0.12). Forty percent of caregivers had a CES-D score of 16 or higher, indicating high level of depressive symptoms. Most caregivers were 'on call' for care for more than 12 hours per day (Table 5.1). The large majority of caregivers could rely on others to take care of their children.

Table 5.1 Characteristics of children with ASDs, caregivers, care situations, and bivariate analyses of CarerQol-VAS scores (One-Way ANOVA test), n=224

Characteristic		%	mean CarerQol-VAS
Child			
Age	<8.4 years	61	7.4±2.0
	≥8.4 years	39	7.3±1.9
Gender	boy	87	7.3±2.0
	girl	13	8.0±1.6
First child	yes	50	7.5±1.7
	no	50	7.3±2.1
HUI-3	<0.7	40	**7.1±1.9**
	≥0.7	60	**7.7±1.8**
QWB-SA	<0.6	52	7.2±1.8
	≥0.6	48	7.6±2.0
Caregiver			
Age	<39.5 years	51	7.6±1.8
	≥39.5 years	49	7.2±2.0
Gender	woman	89	7.5±1.9
	man	11	7.1±2.0
Married	yes	76	7.5±1.9
	no	24	7.1±2.0
Educational level	high school or lower	11	7.8±1.6
	some college	27	7.3±1.9
	college	31	7.4±1.9
	professional or graduate school degree	30	7.3±2.1
Paid work	yes	61	7.5±1.8
	no	39	7.3±2.0
Income	<$20,000	14	7.1±1.9
	$20,000-$35,000	13	7.3±2.1
	$35,000-$60,000	15	7.9±1.8
	$60,000-$100,000	23	7.4±1.9
	>$100,000	36	7.4±1.9
Subjective health (0-100)	<77.6	41	**6.7±1.7**
	≥77.6	59	**7.9±1.8**
EQ-5D score	<0.8	69	**6.9±2.0**
	≥0.8	31	**8.5±1.2**
SF-6D score	<0.7	55	**6.6±1.9**
	≥0.7	45	**8.3±1.5**
CES-D score	<16	60	**8.4±1.2**
	≥16	40	**6.0±2.0**
Care situation			
Hours per day ‘on call’ for caregiving	<8 hours	16	7.6±1.6
	8-12 hours	24	7.7±1.8
	>12 hours	60	7.2±2.0
Other caregivers for care children	no	9	**6.4±2.0**
	yes	91	**7.5±1.9**
Subjective burden			
SRB	<3.6	59	**7.8±1.8**
	≥3.6	41	**6.9±1.9**
FQLS	<100.9	39	**6.2±2.1**
	≥100.9	61	**8.1±1.4**

Note: cut-off points for continuous variables were based on the mean of these variables. Statistically significant differences are in bold ($p<0.05$)

5.3.2 Research objective 1: the impact of caregiving on parents of children with ASDs

Figure 5.1 presents the CarerQol scores. Almost all caregivers of children with ASDs derived fulfilment from caring for their child (97%). Many caregivers experienced problems with combining their care tasks with daily activities (61%). Mental (58%) and physical (52%) health problems and financial problems (56%) were also prevalent. On average, caregivers scored a 7.4 (SD 1.9) on the CarerQol-VAS (Figure 5.1). Table 5.1 shows descriptive statistics of SRB and FQLS. Among caregivers of children with ASDs, the mean SRB score was 36.2 (SD 29.6) and the mean FQLS score was 100.9 (SD 16.1).

5.3.3 Research objective 2: construct validation CarerQol

5.3.3.1 Convergent validity: associations between CarerQol-VAS and CarerQol-7D

Table 5.2 presents Spearman's correlation coefficients of the CarerQol-VAS with the seven burden dimensions of the CarerQol-7D. CarerQol-VAS scores were positively associated with the two positive CarerQol-7D dimensions: fulfilment from caring and support with caring. CarerQol-VAS was negatively associated with the five negative CarerQol-7D dimensions (relational problems, mental health problems, problems with daily activities, financial problems, and physical health problems). These correlation coefficients of the CarerQol-VAS with the CarerQol-7D dimensions had small to high strength of correlation.

Table 5.2 Spearman's correlation coefficients between CarerQol, SRB and FQLS domains, n=201

	CarerQol-VAS			**CarerQol-7D**				
		Fulfilment	**Relational problems**	**Mental health problems**	**Problems with daily activities**	**Financial problems**	**Support**	**Physical health problems**
CarerQol-VAS	1	0.39***	-0.29***	-0.54***	-0.43***	-0.35***	0.20**	-0.50***
SRB	-0.30***	-0.31***	0.30***	0.32***	0.38***	0.21**	-0.05	0.31***
Family Quality of Life Scale								
Family interaction	0.51***	0.29***	-0.27**	-0.34***	-0.35***	-0.23***	0.25***	-0.31***
Parenting	0.41***	0.33***	-0.26***	-0.25***	-0.30***	-0.25***	0.14*	-0.28***
Emotional well-being	0.53***	0.34***	-0.21**	-0.40***	-0.43***	-0.44***	0.36***	-0.40***
Physical/material well-being	0.25***	0.12*	-0.15*	-0.17*	-0.23***	-0.48***	0.13	-0.39***
Disability-related support	0.39***	0.45***	-0.37***	-0.30***	-0.29***	-0.33***	0.19*	-0.25**

Note: * p<0.05, ** p<0.01, *** p <0.001

Multivariate associations of the CarerQol-VAS and CarerQol-7D show that CarerQol-VAS scores were lower among caregivers who derived no or only some fulfilment from caregiving compared to those who derived a lot (Table 5.3). CarerQol-VAS scores were also lower among those experiencing physical or mental health problems. Subgroup analysis of the multivariate associations of the CarerQol-VAS and CarerQol-7D showed that fulfilment was associated with CarerQol-VAS among caregivers with high subjective burden and low family quality of life. Among subgroups of caregivers with low SRB and those with high FQLS scores, support was associated with CarerQol-VAS. Physical health problems and financial problems were associated with CarerQol-VAS among the subgroup of caregivers with high SRB (Table 5.3).

Table 5.3 Regression analyses with CarerQol-VAS as dependent variable among all caregivers and subgroups based on SRB and FQLS scores, standardized coefficients

		Subgroups of caregivers			
		Based on SRB score		**Based on FQLS score**	
CarerQol-7D	**All caregivers (n=209)**	**Below average burden (n=120)**	**Average or higher burden (n=83)**	**Below average family Qol (n=75)**	**Average or higher family Qol (n=120)**
Fulfilment					
no / some (ref. a lot)	-0.23***	-0.15	-0.29**	-0.30**	-0.11
Relational problems					
some / a lot (ref. no)	0.05	0.03	0.11	0.06	0.17
Mental health problems					
some / a lot (ref. no)	-0.28***	-0.31**	-0.24*	-0.42**	-0.28**
Problems with daily activities					
some / a lot (ref. no)	-0.05	-0.12	0.03	0.02	-0.08
Financial problems					
some / a lot (ref. no)	-0.12	-0.04	-0.22*	-0.04	-0.13
Support					
no / some (ref. a lot)	-0.10	-0.23*	0.04	0.05	-0.18*
Physical health problems					
some / a lot (ref. no)	-0.16*	-0.06	-0.24*	-0.16	-0.10
Constant	9.37	9.41	8.97	8.63	9.12
Adjusted R^2	0.32	0.29	0.27	0.26	0.22

Note: * $p<0.05$, ** $p<0.01$, *** $p<0.001$

5.3.3.2 Convergent validity: associations between CarerQol and other instruments of impact of caregiving

Spearman's correlation coefficients of the CarerQol with subjective burden of caregiving (SRB) and family quality of life (FQLS) are displayed in Table 5.2. First, CarerQol-VAS and CarerQol-7D dimension fulfilment were significantly negatively associated with SRB scores and positively with FQLS domain scores.

These correlation coefficients had small to high strength. CarerQol-7D dimension support was significantly associated to four of the five subdomains of FQLS. Relational problems, mental health problems, problems with daily activities, financial problems, and physical health problems of the CarerQol-7D were all positively associated with SRB scores and negatively associated with FQLS domain scores (small to moderate strength of correlation coefficients).

Table 5.4 Mean values of the health of children with ASDs (HUI-3) and caregivers' well-being (CarerQol-VAS), subjective burden (SRB), family quality of life (FQLS) and health (EQ-5D, CES-D) per 'extreme level' of CarerQol-7D, n ranges from 210 to 221 on CarerQol-7D items

CarerQol-7D	HUI-3 child	CarerQol-VAS	SRB	FQLS	EQ-5D caregiver	CES-D caregiver
Fulfilment						
no & some	0.6 [c]	6.6	46.0	93.3 [a]	0.8 [a]	18.2
a lot	0.7	8.0	28.4	106.9	0.9	11.0
Relational problems						
no	0.7 [b]	7.8 [a]	27.7	105.3	0.9 [b]	11.2
some & a lot	0.6	7.0	45.1	95.9	0.8	16.8
Mental health problems						
no	0.7	8.4	23.9	107.3	0.9	7.0
some & a lot	0.6	6.6	44.7	96.3	0.8	18.6
Problems combining daily activities						
no	0.7	8.3	23.9	107.4	0.9	7.9
some & a lot	0.6	6.8	44.2	96.8	0.8	17.4
Financial problems						
no	0.7 [a]	8.0	31.4 [b]	106.9	0.9	10.7
some & a lot	0.6	6.8	40.2	95.5	0.8	16.6
Support						
no & some	0.6	7.1	39.5 [a]	97.5	0.8	15.7
a lot	0.7 [b]	8.4	26.8	110.3	0.9	8.4
Physical health problems						
no	0.9 [a]	8.3	24.6	106.7	0.9	7.8
some & a lot	0.8	6.7	45.5	96.3	0.8	18.6

Note: All differences in mean values were statistically significant at 99.9% C.I., unless otherwise specified
[a] $p<0.1$, [b] $p<0.05$, [c] ns. Results of QWB-SA resemble those of the other indicator of child's health (HUI-3). The results of the other health indicator of caregivers (SF-6D) do not differ from EQ-5D results.

5.3.3.3 Discriminative validity

Investigating differences in outcome scores among 'extreme' groups of caregivers showed that caregivers of children with ASDs that derived a lot of fulfilment or received a lot of support had higher mean general quality of life (CarerQol-VAS) and family quality of life (FQLS) (Table 5.4). Moreover, their child's and their own health were better compared to caregivers with only some or no fulfilment or support. Furthermore, caregivers with a lot of fulfilment or support had lower subjective burden scores (SRB) compared to others. The same results were found for differences in mean values among caregivers without relational, mental health, daily activities, financial or physical health problems compared to those who did experience problems (Table 5.4). These differences in mean values were all statistically significant ($p<0.05$). The only exception was that no differences were found in the level of the child's health among caregivers with or without fulfilment.

5.3.3.4 Clinical validity

Bivariate associations of the CarerQol-VAS scores and child, caregiver and care situation characteristics are presented in Table 5.1. The CarerQol-VAS score was higher among caregivers in relatively good physical or mental health. CarerQol-VAS score was also higher among caregivers of children with relatively good health. Concerning the care situation, the CarerQol-VAS score was higher among caregivers that could rely on others for assistance with care. Multivariate regression analysis shows no statistically significant association of the CarerQol-VAS and background characteristics (results not presented).

5.4 Discussion

Our study had two research objectives: (i) providing insight in the impact of caregiving on parents of children with ASDs and (ii) construct validation of the CarerQol. Concerning the first research objective, as found by others [183-185], children with ASDs in the study sample had relatively poor overall health, with for example, an average HUI-3 score of 0.7 compared to a HUI-3 score of 0.9 among a healthy control group including many children and adolescents in the US [204]. The caregivers of children with ASD in this study predominantly were married mothers in their forties combining care for their child(ren) with a paid job. Thus, given parents' life phase, they often found it difficult to combine care tasks with daily activities such as work, household activities and family life. Moreover, in line with other findings [187, 205, 206], many caregivers of a child with an ASD experienced financial or mental health problems. In the study sample, 40 per cent of parents reported high level of depressive symptoms, which is comparable to levels reported in similar studies of parents of children with ASDs [205, 207]. By contrast, the prevalence of depressive mood has been reported to be lower amongst parents of children with craniofacial malformations [54] or among parents of typically developing children [208]. Despite such difficulties, many parents of children with ASD reported that they derived fulfilment from caring for their child. Moreover, parents described themselves to be fairly happy, scoring more than a 7 on a 0 to 10 scale of happiness. This result is consistent with previous findings (chapters 2 and 3, [54, 110]).

Before discussing the results of the second research question on construct validation of the CarerQol, some study limitations need to be addressed. First, some caution is needed in generalizing the results to all families of children with ASDs given some limitations regarding the study sample and data collection. For example, in the study sample of children with ASDs boys were somewhat overrepresented with a boy: girl ratio of 6.7 to 1, while this ratio is 4.6 to 1 in the United States [209]. Moreover, the study sample seems to include relatively many highly educated working mothers. In addition, it should be stressed that the results might have been affected by the use of two specific ATN sites, in Arkansas and New York. In general, these ATN sites differ in terms of the availability of formal and informal care resources given wealth and cultural differences between northern and southern states in the US. Moreover, the ATN site in New York treats children with more severe disorders.

Although no differences were detected in the health status of children between ATN sites, parents from the New York ATN site experienced higher subjective burden of caring (SRB) than parents from the Arkansas' ATN site. Scores on other outcome variables used in this study did not differ between parents of these two ATN sites. Given the focus on construct validation, this finding is not considered to be problematic. Another limitation in the data collection concerned the use of parent-proxy reports. Although these parent-proxy reports are often used in research on children with ASDs due to concerns about the reliability of self-reports from children who often have severe cognitive and communication problems [184], this might have biased the results. However, the impact of this seems relatively small in this study given that the analyses mostly concerned parent variables. Finally, in this study none of the background characteristics of the child with ASDs, parent or care situation were significantly related to caregivers' well-being after adjusting for subjective burden dimensions. Given these limitations, it would be interesting in future studies to collect additional characteristics of the children and their disorders, such as clinical assessments of the severity of the disorder, in order to explain the impact of caregiving on their parents more thoroughly. Furthermore, replication of this study in a larger sample from a broader range of ATN sites is advised.

Concerning the second research question on construct validation of the CarerQol, results of convergent and discriminative validity tests showed that well-being, deriving fulfilment and receiving support among caregivers of children with ASDs were all associated with higher family quality of life and lower subjective burden of caring. By contrast, having relational, mental health, daily activity, financial or physical health problems were associated with more subjective burden of caring, lower family quality of life and lower overall well-being. When considering the influence of subjective burden dimensions on caregivers' well-being, especially fulfilment and mental and physical health problems were related to the well-being of parents of children with ASDs. Moreover, subgroup analyses showed interesting insights into the various associations between caregivers' well-being and subjective burden dimensions. While some aspects of subjective burden, such as mental health, negatively influenced well-being among all subgroups of caregivers, financial problems were especially relevant to the well-being of caregivers experiencing high burden. Additionally, fulfilment especially influenced well-being among caregivers in a relatively bad situation (i.e., high subjective burden or low family quality of life), while receiving support was particularly relevant to the well-being of caregivers in a relatively good position (i.e., low subjective burden or high family quality of life). Other construct validation tests of the CarerQol instrument also show the diversity of problems that affect well-being among caregivers (chapters 2 and 4); however, whether the influence of the caring problems found in this study are only specific for the care situation of children with an ASD, or apply to children with other care needs as well, is not yet clear. Further research into associates of well-being of parents of children suffering from other disorders or illnesses is needed. Overall, clinical validation results supported the ability of the CarerQol to accurately measure the impact of caregiving, as most background characteristics of caregivers, care recipients and care situations associated with caregivers' well-being in this study were consistent with findings from previous CarerQol validation studies (chapters 2 and 3, [54, 110]). Lastly, a note should be made on the support dimension of the CarerQol. While convergent and discriminative validity tests show inconsistent results in terms of statistical significance, all results seem to imply that support has a positive effect on parents of children with ASDs. This is also found by others [186, 189, 191, 207, 210]. However, receiving support also has some downsides. For example, different support resources, such as social networks, professional health care or special educational services, are often used for children with special health needs which require some kind of coordination [186, 211]. Moreover, support is not always considered as helpful by parents of children with ASDs [186]. Hence, support could increase burden when problems arise between caregivers, family and professionals, such as disagreement or communication difficulties [160, 189].

5.5 Conclusions

This study showed that while many parents in the study sample derived fulfilment from providing care for their child with an ASD, this caregiving affected their own lives considerably and in a variety of ways. Parents often experienced problems with combining care with other daily activities, had financial problems or suffered from depressive symptoms. In addition, this study supported construct validity of the CarerQol in a population of caregivers for children with ASDs. Further research in larger, representative samples of parents providing informal care to children with ASDs or other health problems is needed before these results can be generalized. Notwithstanding, this study showed that the CarerQol can be used to validly assess the impact of caregiving on parents of children with ASDs in the study sample. The CarerQol thus seems to provide information that is relevant for evaluations of treatment interventions for children with ASDs or for support interventions for their informal caregivers.

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CHAPTER

6

Measuring caregiver outcomes in palliative care: A construct validation study of two instruments for use in economic evaluations

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Abstract

Purpose

Providing care to patients nearing the end of life can place a considerable burden on caregivers. Hence, policy decisions on interventions in palliative care should be guided by information on this burden. This study investigates construct validation of two preference-based caregiver outcome instruments suitable for economic evaluations: the Carer Experience Scale (CES) and the Care-related Quality of Life (CarerQol) instrument. Moreover, this study reports caregiver experiences in end-of-life care.

Methods

Data was collected with written questionnaires among caregivers of patients receiving palliative care services in the Southern metropolitan area of Adelaide, South Australia. The effect of caregiving on caregivers was measured with the CES, CarerQol, Process Utility (PU) and Caregiver Strain Index (CSI). Convergent, discriminative, and clinical validity were studied.

Results

As hypothesized, higher negative effect of caregiving measured on the CES was associated with higher negative effect on the CarerQol. Both the CES and CarerQol were associated in the expected positive direction with less strain from caregiving (CSI), more positive care experiences and more process utility from caring. More negative caring experiences were found the lower care recipients' and caregivers' health status, and the longer the duration of caregiving.

Conclusions

The findings suggest that the CES and CarerQol validly assess the effect of caregiving on caregivers in end-of-life care for use in economic evaluations. Economic evaluations in end-of-life care should attempt to incorporate such instruments to provide a more holistic assessment of the true impact of interventions, especially where family and friends are heavily involved in caregiving.

6.1 Introduction

Family and friends are often central in the care for terminally ill individuals receiving palliative care services. These informal caregivers may assist with many aspects of patients' lives, whilst at the same time, they are also affected by the fact that their loved one is seriously ill [50, 53]. Furthermore, in palliative care caregivers also need to cope with their loved-ones nearing the end of life [212-214]. Caregivers in palliative care typically provide emotional support, perform household tasks, provide personal care, assist with administrating medication, fulfil administrative duties and provide round-the-clock physical care [212, 215]. Hence, many caregivers providing end-of-life care often feel stressed by caregiving [216]. Often, they experience problems, such as anxiety, depressive mood, fatigue, physical health problems, reduced sleep, difficulties maintaining paid work and financial burden [212, 215-218], sometimes even to the point that they may choose not to take on a similar role again in the future [219]. Furthermore, many caregivers report unmet needs, such as lack of emotional support [220]. Despite this, evidence indicates that caregivers may also experience positive benefits associated with their role in end-of-life care [221, 222], such as learning better to cope with problems in life and gaining more appreciation for others [223].

In the literature, little is known about what causes and influences caring experiences in end-of-life care [224]. That is, most studies in palliative care only focus on patients [224, 225]. In end-of-life research, caregivers often serve as proxies for patients who are too ill to respond to questions on, for example, satisfaction or experience with specialised palliative care (e.g., [226-228]). Alternatively, studies that do report caregiver experiences in palliative care mainly are descriptive and hence fail to unravel associated factors of these experiences. These studies typically focus only on specific aspects of the caregiving situation. For example, studies report the number of hours of caring per week, the type of care tasks performed [212, 215] or the health status of end-of-life caregivers [218, 229]. However, only focussing on such specific aspects of the care situation can be misleading. For example, reports of caregiving burden based on caregivers' time input do not necessarily match strain felt by caregivers themselves [25, 29, 34, 100]. Moreover, outcomes reported in terms of caregivers' health status or health-related quality of life, may be incomplete, because, as previously described [212, 215-218], health issues are only one of the many consequences caregivers might experience in the palliative care context. Hence, instruments measuring caregiver-reported outcomes [224, 230] and covering the complete range of the caregiving experience are needed for documenting the impact of caregiving on carers. All possible effects of caregiving could, at least in theory, be captured in terms of well-being of caregivers. At the same time, however, it is highly likely that caregivers' well-being will also be influenced by factors of life beyond caregiving, such as wealth of caregivers [53, 103]. Instruments that focus more directly on the caregiving experience are subjective burden instruments [26, 104, 105, 161]. These instruments record problems of different aspects of caring, such as financial problems or difficulties performing social activities [25, 29]. Some subjective burden instruments are disease or problem specific, such as the Family Caregiver Medication Administration Hassles Scale [231]. Other instruments are applicable in different caregiver populations, such as the Caregiver Strain Index [107], Caregiver Reaction Assessment [215, 217, 232, 233], Revised Caregiving Appraisal Scale [234], Family Strain Questionnaire [218, 235] or Zarit Burden Interview [106, 216].

Whilst these subjective burden instruments are informative of the experiences of caregiving and, as such, may be usefully applied in palliative care studies to report the strain of caring in caregivers, they are not suitable for application in studies recording the effectiveness or value for money of interventions for palliative patients or their caregivers. In general, in health care, reimbursement decisions are increasingly being guided by cost-effectiveness information [114, 236]. At this moment, informal care is typically ignored when cost-effectiveness of health care interventions is calculated in economic evaluations. In care situations such as palliative care where family and friends provide a large part of the care for people in need of care [16, 61] ignoring informal care in economic evaluations might bias cost-effectiveness research. Consequently, policy makers may be ill informed and choose interventions that do not optimally benefit society as a whole [25, 75, 79]. Economic evaluation studies require a single score reflecting the overall effect of caring on caregivers to facilitate comparisons of different interventions. Ideally, this overall score should be based on both problems and positive experiences of caring [28, 222]. Moreover, the overall score needs to be preference weighted reflecting the relative importance of these dimensions of caregiving, because some problems of caring might be more burdensome than others [34].

Two caregiver outcome instruments, the Carer Experience Scale (CES; [108, 113]) and the Care-related Quality of Life (CarerQol; [110]) instrument, satisfy these requirements for inclusion in economic evaluations. Both instruments were specifically designed for measuring the overall effect of caregiving on caregivers for inclusion in economic evaluations (chapter 7, [108, 110, 113]) . The CES (Figure A6.1) measures care-related welfare and consists of six subjective burden dimensions of caregiving: (i) activities outside caring, (ii) support from family and friends (social support), (iii) assistance from organizations and the government (institutional support), (iv) fulfilment from caring, (v) control over the caring, and (vi) getting on with the care recipient [108]. Relative weights attached to each of the six care dimensions are aggregated to provide a preference-based overall score of caring experiences (0 'bottom state' - 100 'top state'). A scoring algorithm, based upon the preferences of caregivers of older people in the UK, was used to calculate this score [113]. This scoring system is based on preferences for caregiving dimensions described with the six dimensions of the CES derived with a best-worst scaling (BWS) experiment. More detailed information on the BWS methodology and the scoring itself can be found in Al-Janabi, Flynn & Coast (2011). The CarerQol (Figure A6.2) consists of two separate parts: a measure of subjective burden (CarerQol-7D) and of well-being (CarerQol-VAS) [110, 162, 237]. The CarerQol-7D asks respondents for a description of their caregiving situation based on two positive (fulfilment and support) and five negative (relational problems, mental health problems, problems combining daily activities with care, financial problems, and physical health problems) caregiving dimensions. The seven burden dimensions of the CarerQol-7D can be aggregated and weighted by their severity with a tariff. This tariff is based on preferences for different caregiving situations described by the seven dimensions of the CarerQol-7D. These preferences were obtained with a discrete choice experiment among the general public in the Netherlands (chapter 7). The tariff can be found in chapters 7 and 8 and can be used to calculate an overall score for the CarerQol-7D that ranges from 0 (worst informal care situation) to 100 (best informal care situation). The other part of the CarerQol instrument is the CarerQol-VAS. The CarerQol-VAS provides a valuation of informal care in terms of well-being by measuring happiness of caregivers on a horizontal visual-analogue scale (VAS) ranging from completely unhappy (=0) to completely happy (=10). The CarerQol-7D and the CarerQol-VAS both provide an overall score of the impact of caregiving on caregivers. Both the CarerQol-7D overall score and the CarerQol-VAS score can, separately, be used as outcomes in caregivers in economic evaluations. However, the concept that the CarerQol-VAS values, i.e., well-being, might, as previously discussed, be too broad in the context of informal care. The CarerQol-7D overall score tries to overcome this problem by focusing more directly on problems of caregivers experienced in the caregiving context.

When the term 'CarerQol' is used in this chapter, it refers to the complete instrument consisting of the two separate parts of CarerQol-VAS and CarerQol-7D.

Construct validation results of the CES and CarerQol are encouraging. Evidence for the CES comes from a UK general population sample of caregivers [111]. Goranitis, Coast & Al-Janabi (2014) studied construct validation of the CES by focussing on the association between the CES and diverse contextual variables. They found that, as expected, both characteristics of the care situation, care recipient and caregiver were associated with single CES dimensions and the overall CES score. For example, they found that the dimension activities was associated with the type of caregiving tasks performed, whether the caregiver was the main carer, hours of caregiving and duration of caregiving. Furthermore, as anticipated, they found that the overall CES score was also associated with these care situation variables. In addition, CES scores were associated with caregivers' and care recipients' characteristics, such as their health. The study of Goranitis, Coast and Al-Janabi (2014) further strengthens the construct validation of the CES by showing that nearly three in four anticipated hypotheses about the association between the CES and contextual variables were statistically significant in their sample of caregivers. Construct validation studies of the CarerQol were conducted in samples of caregivers that mainly cared for adult care recipients with a chronic disease or health problems due to aging or for children with special health needs (chapters 2, 3 and 4, [110]). The CarerQol seems a valid instrument of the impact of caring on caregivers in these caregiver populations, because the CarerQol is associated with important contextual variables, for example hours of caregiving, relationship between caregiver and care recipient, and health status of caregiver and care recipient in these studies. Moreover, results of these validation studies show that the CarerQol is associated with other instruments measuring the same construct as the CarerQol, for example the Caregiver Strain Index (CSI; [107]), Self-Rated Burden scale (SRB; [29, 105]) and Process Utility (PU; [28]), in the expected direction. That is, more negative caregiving experiences on the CarerQol were associated with more subjective burden from caregiving on the CSI, SRB and PU.

This study has two general aims. First, to date, little is known on how the overall effect of caring on caregivers can be measured in the palliative care setting. This study, therefore, investigates construct validity of the CES and the CarerQol in end-of-life care. In this study, an extensive dataset of caregivers caring for a loved one with a disease in its terminal phase was used. This dataset contains several instruments that measure the effect of caregiving amongst these caregivers. This provides the unique opportunity to study construct validation of the CES and the CarerQol and to investigate caregiver experiences in end-of-life care. A second aim of this study is to assess the effect of caregiving on caregivers in end-of-life care. More insight into these caring experiences in palliative care is important, because this specific group of caregivers is confronted with a range of care situations given diverse trajectories of mental and functional decline of palliative care patients and different access and uptake of formal care services [225].

6.2 Methods

6.2.1 Data

Written questionnaires (N=300) were distributed to a consecutive cohort of caregivers of patients attached to Southern Adelaide Palliative Services, South Australia, by community nurses as they did home visits during a four month time period, January to April 2011. For people whose next visit was not due for some time, the questionnaire was posted with a note from their community nurse. Permission was not sought from patients at any time. Return of the questionnaire was taken as implied consent of the caregivers. A total of 99 questionnaires were returned of which two were excluded[2], resulting in 97 participating caregivers (32% response rate).

6.2.2 Questionnaire

The questionnaire included information on (i) the informal care situation, (ii) background characteristics of the caregiver, and (iii) background characteristics of the care recipient (Table 6.1). Characteristics of the informal care situation consisted of information on the relationship of caregiver and care recipient (partner, parent, other family member, friend/neighbour), whether the caregiver lives with the care recipient, whether the caregiver is the main caregiver, the number of years that the caregiver has been providing care, the type of care tasks (help with daily living, housework, organisation, other tasks), the number of hours spent on caring and the type of motivation to start providing informal care (answering categories: duty to care, closest person, free choice, no-one else to provide care, no money for paid care).

Background characteristics of the caregiver were age, gender, age when leaving full-time education, employment status (full-time, part-time, self-employed, no paid employment), ethnicity (Australian/European, Aboriginal/Torres Strait Islander, Asian, African, Pacific Islander). Caregivers' health status was measured with the EQ-5D instrument [122] using EQ-5D weights for Australia to calculate health-related quality of life of caregivers [238]. Information was collected on the palliative care patient's gender, age, and type of health problem (problems with physical health, mental health, intellectual health or a combination of these health problems).

Moreover, the questionnaire measured the effect of caregiving on the caregiver with four subjective burden instruments: the CES, CarerQol instrument, PU, and the Expanded Caregiver Strain Index (CSI+). PU (Figure A6.3) concerns the value people attach to the process of providing informal care [28]. Two questions are used to calculate PU. The first question measures happiness of caregivers in their current situation (similar to CarerQol-VAS). The second question instructs respondents to imagine that the care would be handed over to a person of the care recipients and caregivers' choice. After that, caregivers are asked how happy they would be in this situation using a horizontal VAS (0 'completely unhappy' – 10 'completely happy'). PU (range -10 to 10) is the difference on these two happiness questions and a higher score indicates a higher level of process utility of caring. PU has been applied in several studies and is associated with care situation characteristics, such as caregiver health or time spent on caregiving, and with positive or negative caregiving experiences of caregivers (chapters 2 and 4, [28, 33, 110]). The CSI (Figure A6.4) [107] measures the strain of caregiving experienced by caregivers by listing 13 items referring to stressors in caring with two choice options: yes (score 1) or no (score 0). The CSI provides an unweighted summary score (referred in this chapter as sum score negative items CSI+; range 0-13) whereby a higher score indicates higher caregiving strain. Caregivers are considered to experience substantial strain if their score is 7 or higher [107]. The CSI is often used in research to measure strain from caregiving and studies show good psychometric properties of the instrument concerning feasibility, reliability and construct validity [105, 107, 110, 239-241]. For example, construct validity of the CSI is supported by correlations with both other instruments measuring caregiving experiences and with patient and caregiver characteristics, such as their health status [105, 240]. In this questionnaire, five statements on positive aspects of caregiving were added to the original CSI instrument, forming the CSI+ as proposed by Al-Janabi et al. (2010) (Figure A6.4).

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[2] One respondent returned an empty questionnaire and the other did not define themselves as a caregiver.

The positive items of the CSI+ have two answering categories (yes; score 1 /no; score 0), that can be summed up to an overall score with a higher score meaning more positive experiences from caring (sum score positive items CSI+; range 0-5). The study of Al-Janabi et al. (2010) presented evidence of construct validity for the CSI+, and showed that, for example, the positive items of the CSI+ were correlated with other subjective burden instruments and with the health-related quality of life of patients and caregivers.

6.2.3 Statistical analyses first research question: construct validation

Construct validation of the CES and the CarerQol was studied with convergent, discriminative, and clinical validity tests [98].

6.2.3.1 Convergent validity

The focus of convergent validity is on testing whether a construct underlying an instrument resembles that of other instruments with the same subject of measurement [98]. In this study, convergent validity of the CES and the CarerQol was assessed by studying the association between these instruments. A priori, a high degree of correlation (Spearman's rho: 0.5-0.7) between the CES and the CarerQol was expected, because both instruments are similar in content and aim [110, 112].

Secondly, the association of CES and CarerQol with the other instruments of the effect of caregiving (PU, sum score of the negative CSI+ items, and sum score positive items CSI+) was analysed. It was hypothesized, based on results of previous validation studies (chapters 2 and 4, [110]), that more negative caring experiences on the CES and CarerQol were associated with higher sum scores of the negative items of the CSI+ and lower PU and positive items CSI+ sum scores.

Thirdly, associations between single dimensions of the CES and the CarerQol-7D were investigated. Moreover, association between the CES and a selection of dimensions from the comprehensive list of the CSI+ were analysed. The seven CarerQol-7D dimensions were tested in the same way. Associations between CES dimension 'activities' and CarerQol-7D dimension 'problems daily activities' were hypothesized. Associations between these CES and CarerQol-7D dimensions and CSI+ dimensions 'enough time for self' and 'other demands on time' were also expected. CES dimensions 'social support' and 'institutional support' were expected to be associated with CarerQol-7D dimension 'support'. Further, associations between CES dimension 'fulfilment', CarerQol-7D's 'fulfilment', and CSI+ dimension 'happy to care' were hypothesized. CES dimension 'getting on' was expected to be associated with CarerQol-7D's 'relational problems'. Furthermore, an association between CarerQol-7D's 'financial problems' and CSI+ dimension 'financial strain' was hypothesized. Lastly, a priori, an association between CarerQol-7D dimension 'physical health problems' and CSI+'s 'physical strain' was expected. Hypotheses were formulated using the authors' judgment.

All convergent validity tests were analysed with Spearman's correlation coefficients. The strength of correlation coefficients was assessed as follows: <0.1 trivial; 0.1–0.3 small; 0.3–0.5 moderate; 0.5–0.7 high; 0.7–0.9 very high; >0.9 nearly perfect [174].

6.2.3.2 Discriminative validity

Discriminative validity focuses on the ability of an instrument to differentiate between relevant groups of respondents [98]. To study discriminative validity of the CES and CarerQol, 15 pairs of different groups of caregivers in terms of the strain experienced from caregiving were constructed. These groups were specified using the instruments recording caregiving experiences included in this chapter: CES (six pairs of caregivers; few or many problems with other activities, a lot or little social support, a lot or little institutional support, mostly or sometimes/rarely fulfilment, control over most or few caring aspects, mostly or sometimes/rarely getting on with care recipient), CarerQol-7D (seven pairs of caregivers; a lot or no/some fulfilment, no or some/a lot of relational problems, no or some/a lot of mental health problems, no or a lot of problems with daily activities, no or a lot of financial problems, a lot or no support with caring, no or a lot of physical health problems), CSI (one pair of caregivers; lower or equal/higher than sum score of the negative items of the CSI+ of 7) and PU (one pair of caregivers; negative or positive PU). We hypothesized finding higher negative effect of caring scores among caregivers in negative extreme groups. Among caregivers in these groups, mean values of CES, CarerQol-7D overall score, CarerQol-VAS, PU, sum score of the negative items of the CSI+ and sum score positive items CSI+ were compared. The differences in mean values were tested with Student's t-tests. Non-parametric statistical tests (Wilcoxon's test) were used for validation of these results.

6.2.3.3 Clinical validity

Clinical validity is closely related to convergent and discriminative validity tests. Clinical validity tests focuses not on whether the scale is associated to other instruments with the same underlying construct as convergent and discriminative validity do, but rather focuses on association of the measured construct with important explanatory variables. Clinical validity assesses whether respondents with differences in caring experiences have different scores on a scale [98]. In this study, clinical validity was analysed with bivariate associations of both the CES and CarerQol-7D overall score with background characteristics of the caregiver, palliative care patient, and informal care situation. Spearman's correlation coefficients were used for continuous variables and Student's t-tests for categorical variables. For validation of these latter results, Wilcoxon tests were used. Given results of previous CES or CarerQol validation studies (chapters 2 and 3, [110, 111, 112]), in the context of end-of-life care associations of the CES and CarerQol-7D overall scores with the type of relationship between the caregiver and patient were expected. Furthermore, a higher negative effect of caregiving among caregivers or patients with more health problems was hypothesized. The other background characteristics are used for exploratory testing of factors associated with the caregiving experience measured with the CES and the CarerQol-7D overall score in the specific setting of palliative care.

6.2.4 Statistical analyses second research question: effect of caregiving on caregivers in end-of-life care.

Descriptive statistics in percentages and means (SD) were calculated for care situation, caregiver and palliative care patient characteristics. All statistical analyses were performed with Stata® version 11.0 (StataCorp LP).

Table 6.1 Descriptives of informal care situation, caregiver and care recipient characteristics in percentages or mean (SD), n=97

Characteristic	Statistic		Value
Care situation	Relationship [b]	partner	59%
		parent	29%
		other family member	6%
		friend or neighbour	4%
	Main caregiver		90%
	Sharing household		82%
	Number of years		3.9 (5.7)
	Number of hours per week		61.2 (45.4)
	Tasks [b, d]	help with daily living	70%
		housework	75%
		organisation	84%
		other	79%
	Motivation to care [c, d]	duty to care	55%
		closest person	80%
		free choice	59%
		no-one else	43%
		no money for paid care	20%
Caregiver	Age		62.3 (11.9)
	Gender [b]	female	71%
		male	27%
	Educational level [b]	<=16 years	36%
		17 or 18 years	21%
		19 years or over	41%
		still full time education	0%
	Employment [b]	full-time	12%
		part-time	16%
		self-employed	9%
		no employment	60%
	Ethnicity [a]	Australian/European	98%
		Aboriginal/Torres Strait Islander	0%
		Asian	1%
		African	0%
		Pacific Islander	0%
	EQ-5D score		0.7 (0.2)
Palliative care patient	Age		72.3 (11.9)
	Gender [b]	female	46%
		male	52%
	Health problem [b]	only physical	56%
		only mental	0%
		only intellectual	0%
		physical and mental	23%
		physical and intellectual	6%
		mental and intellectual	0%
		physical, mental and intellectual	12%
		no health problems	1%

[a] 1% missing values, [b] 2% missing values, [c] 3% missing values, [d] percentages add up to >100%, because respondents could choose more than one category

6.3 Results

6.3.1 First research question: construct validation

6.3.1.1 Convergent validity

Table 6.2 presents the Spearman's correlation coefficients of the CES, the CarerQol-7D overall score, and CarerQol-VAS with PU, sum score of the 13 negative CSI+ items and sum score of the five positive items of the CSI+. CES scores were positively associated with CarerQol-7D overall scores (high strength of correlation coefficients) and with CarerQol-VAS scores (moderate strength). CES scores had a positive association with PU scores (moderate strength). CES scores were positively associated with the sum score of positive items of the CSI+ (high strength) and negatively with the sum score of the negative CSI+ items (moderate strength).

CarerQol-7D overall scores were positively associated with CarerQol-VAS scores. The CarerQol-7D overall score had a positive association with PU and with the sum score of the positive items of the CSI+. A negative association was found for CarerQol-7D overall score with the CSI+ negative item sum score. All associations in Table 6.2 had the expected direction and were strong, except the association between CarerQol-VAS and the sum score positive items CSI+ that had moderate strength.

Table 6.2 Spearman's rho of Carer Experience Scale (CES), CarerQol instrument (CarerQol-7D & CarerQol-VAS), Process Utility (PU) and Expanded Caregiver Strain Index (CSI+; sum score negative items CSI+ & sum score positive items CSI+), n ranges from 75 to 95

	CES	CarerQol-7D overall score	CarerQol-VAS	PU	sum score negative items CSI+
CES	-				
CarerQol-7D overall score	0.57	-			
CarerQol-VAS	0.42	0.60	-		
PU	0.41	0.62	0.70[a]	-	
sum score negative items CSI+	-0.46	-0.67	-0.59	-0.47	-
sum score positive items CSI+	0.60	0.55	0.45	0.44	-0.47

Note: All correlation coefficients are significant at $p<0.001$

[a] Important to note is that the association between the CarerQol-VAS and PU should be interpreted with caution. A high correlation may be expected, given that PU is calculated using CarerQol-VAS scores.

The associations of the single dimensions of the CES, the CarerQol-7D and a selection of single CSI+ dimensions are presented in Table 6.3. All hypothesized associations of the CES and the CarerQol-7D dimensions were statistically significant and had moderate strength, except the association between the CES and CarerQol-7D dimensions covering 'fulfilment' that had very high strength. Moreover, almost all a priori expected associations of CES and CarerQol-7D dimensions with single CSI+ dimensions were statistically significant. Most of these associations had moderate strength, however the CES dimension 'activities' was strongly associated with CSI+ 'enough time for oneself' and was not statistically significantly associated with CSI+ item 'other demands on time'.

Table 6.3 Associations of single dimensions of the Carer Experience Scale (CES), CarerQol-7D and a selection of the dimensions of the Expanded Caregiver Strain Index (CSI+) using Spearman's correlation coefficients, n ranges from 89 to 97, hypothesized associations in bold

	CES						CSI+				
CarerQol-7D	**Activities**	**Social support**	**Institutional support**	**Fulfilment**	**Control**	**Getting on**	**Physical strain**	**Enough time for self**	**Other demands on time**	**Happy to care**	**Financial strain**
Fulfilment	0.01	0.24	0.07	**0.83*****	0.32**	0.38***	-0.05	0.00	-0.01	**0.33****	-0.06
Relational problems	-0.05	-0.26*	0.00	-0.41***	-0.24*	**-0.48*****	0.12	0.12	0.16	-0.31**	0.15
Mental health problems	0.19	-0.30**	-0.15	-0.09	-0.12	-0.08	0.29**	-0.23*	0.27**	-0.05	0.42***
Problems daily activities	**-0.39*****	-0.30**	-0.04	-0.22*	-0.13	-0.20	0.50***	**-0.46*****	**0.36*****	-0.17	0.30**
Financial problems	-0.10	-0.18	-0.13	-0.09	0.00	-0.07	0.31**	-0.14	0.14	-0.18	**0.46*****
Support	0.17	**0.48*****	**0.30*****	0.31**	0.19	0.29**	-0.08	0.14	-0.13	0.07	-0.27*
Physical health problems	-0.37***	-0.30**	-0.11	-0.08	0.03	-0.09	**0.41*****	-0.32**	0.14	-0.05	0.50***
CSI+											
Physical strain	-0.27**	-0.12	0.03	-0.07	-0.03	-0.21					
Enough time for self	**0.55*****	0.12	0.10	0.05	-0.03	0.05					
Other demands on time	**-0.03**	-0.09	-0.05	-0.11	-0.03	-0.05					
Happy to care	-0.08	0.04	0.00	**0.39*****	0.21*	0.41***					
Financial strain	-0.27*	-0.28**	-0.26*	-0.16	0.04	-0.07					

Note: CES dimension 'control' and CarerQol-7D dimension 'mental health problems' are not presented, because no hypotheses were formulated for these dimensions.
Only CSI+ dimensions are presented that were hyppothesized to be associated with single CES or CarerQol-7D dimensions.
* $p<0.05$, ** $p<0.01$, *** $p<0.001$

6.3.1.2. Discriminative validity

Table 6.4 compares caregivers that derived fulfilment, support, or positive process utility from caring to caregivers that did not experience these positive aspects of caring. Moreover, differences among caregivers experiencing problems or substantial strain from caring and among caregivers without these problems or strain are presented in Table 6.4.

Table 6.4 Mean values of Carer Experience Scale (CES), CarerQol instrument (CarerQol-7D & CarerQol-VAS), Process Utility (PU) and Expanded Caregiver Strain Index (CSI+) per 'extreme level' of CES, CarerQol-7D, PU, sum score negative items CSI+ and sum score positive items CSI+, n ranges from 79 to 96

Carer Experience Scale		CES	CarerQol-7D overall score	CarerQol-VAS	PU	sum score negative items CSI+	sum score positive items CSI+
Activities	most	-	**82.6**	**6.8**	**3.2**	**4.1**	**4.4**
	few	-	**68.6**	**5**	**0.3**	**7.1**	**3.7**
Social support	a lot	-	**83.6**	6	2.2	**4.7**	**4.3**
	little	-	**61.1**	5.2	0.4	**7.4**	**3.5**
Institutional support	a lot	-	78.6[a]	6.1	1.8	5.5	**4.4**
	little	-	71.8	5.7	1.2	6.4	**3.9**
Fulfilment	mostly	-	**79.1**	**6.1**	**2.6**	**5.2**	**4.3**
	sometimes/rarely	-	**66.3**	**5.3**	**-0.7**	**7.2**	**3.6**
Control	most aspects	-	**77.3**	6	**2.6**	5.9	4.2[a]
	some/few aspects	-	**71.3**	5.5	**-0.7**	5.9	3.9
Getting on	mostly	-	**76.8**	**6**	**1.9**	5.6[a]	**4.3**
	sometimes/rarely	-	**60.3**	**4.6**	**-1.9**	7.8	**2.8**
CarerQol-7D		CES	CarerQol-7D overall score	CarerQol-VAS	PU	sum score negative items CSI+	sum score positive items CSI+
Fulfilment	no/some	**66.4**	-	5.4[a]	**-0.4**	6.7[a]	**3.7**
	a lot	**77**	-	6.1	**2.7**	5.3	**4.3**
Relational problems	no	**76.7**	-	6	**2.1**	**4.9**	**4.2**
	some/a lot	**67.7**	-	5.5	**0.3**	**7.4**	**3.8**
Mental health problems	no	**77.1**	-	**7.1**	**4.1**	**3.5**	**4.4**
	a lot	**56.3**	-	**4.2**	**-3.8**	**8.5**	**3.5**
Problems daily activities	no	**80.7**	-	**6.9**	**4.5**	**2.6**	**4.7**
	a lot	**58.4**	-	**4.4**	**-3.8**	**9.2**	**3.2**
Financial problems	no	**74.6**	-	**6.1**	**2.0**	**4.7**	**4.2**
	a lot	**62.9**	-	**4.7**	**0.0**	**9.5**	**3.5**
Support	no	**57.9**	-	**4.6**	**-1.1**	5.1	**3.3**
	a lot	**85**	-	**6.7**	**4.0**	4.6	**4.5**
Physical health problems	no	**79.6**	-	**6.9**	**2.8**	**3.9**	**4.5**
	a lot	**63.3**	-	**4.3**	**-2.5**	**9**	**3.5**
Negative items CSI+		CES	CarerQol-7D overall score	CarerQol-VAS	PU	sum score negative items CSI+	sum score positive items CSI+
Sum score negative items CSI+ <7		**78.2**	**82.1**	**6.6**	**3.0**	-	**4.4**
Sum score negative items CSI+ >=7		**66.4**	**65.2**	**4.8**	**-0.9**	-	**3.6**
Process Utility		CES	CarerQol-7D overall score	CarerQol-VAS	PU	sum score negative items CSI+	sum score positive items CSI+
Positive process utility		**77.7**	**80.5**	**6.7**	-	**4.7**	**4.3**
Negative process utility		**64.4**	**62.7**	**4.5**	-	**7.5**	**3.5**

Note: numbers in bold are statistically significant at 95% C.I. (t-test presented, Wilcoxon test as validation). Categories of dimensions of CES/ CarerQol-7D were merged if the number of respondents was too small in one of the categories. If subgroups were constructed using the instrument itself, mean values of this instrument were not computed.[a] significant at 90 % C.I.

The positive extreme groups of caregivers had significantly higher CES, CarerQol-7D overall and CarerQol-VAS scores than caregivers in the negative extreme groups. These differences were statistically significant, with the exception of the difference in CES and CarerQol-7D scores among caregivers that did or did not receive institutional support (CES dimension) (p<0.1.). Moreover, the CarerQol-VAS differences were not statistically significant for caregivers with or without social or institutional support (CES), fulfilment from caring (CarerQol-7D), control over the care situation (CES) or relational problems with the care recipient (CES). PU and sum score of the positive items of the CSI+ were higher and CSI+ negative items sum score was lower among caregivers in the positive extreme groups. Moreover, PU scores did not differ among extreme groups that differed on the CES dimensions social or institutional support. Further, mean sum scores of the negative items of the CSI+ were alike among caregivers that did or did not derive fulfilment (CarerQol-7D), receive support (CarerQol-7D) or institutional support (CES), have control over the care situation (CES) or get on with the care recipient (CES).

6.3.1.3 Clinical validity

Bivariate associations of CES and CarerQol-7D overall score with background characteristics of caregiver, care recipient and care situation are presented in Table 6.5. First, the number of years that caregivers had been providing care was negatively associated with CarerQol-7D overall scores. Moreover, caregivers that experienced better health had higher CES and CarerQol-7D overall scores. Furthermore, caregivers of patients that suffered only from physical health problems had higher CES and CarerQol-7D overall scores than caregivers of patients with mental or intellectual health problems or a combination of health problems.

6.3.2 Second research question: effect of caregiving on caregivers in end-of-life care

Descriptive statistics of the CES, the CarerQol and the other subjective burden instruments are presented in Table 6.6. On a scale from 0 to 100, caregivers of palliative care patients had an average CES score of 72.5 and an average CarerQol-7D overall score of 73.5. The mean CarerQol-VAS score (0-10) was 5.8. Caregivers reported problems with performing activities outside caring (73% on CES, 74% on CarerQol-7D) and with their own mental or physical health (70% on both instruments). The large majority of caregivers derived fulfilment from providing care (91% on CES, 92% on CarerQol-7D). More than four in five caregivers received support from their family or friends with the care tasks (84% on CES, 87% on CarerQol-7D). Assistance from organisations or the government was provided to around three in four caregivers (74%).

The mean PU score (-10 – 10) was 1.4. More than half of caregivers derived positive process utility (56%). Around one in three had negative process utility (36%). The mean CSI+ negative items sum score was 5.9 (0-13) and 44 per cent of caregivers experienced substantial strain from caring. Many caregivers indicated that providing care for their loved one was confining (63%), that they had to make changes in their personal plans (72%) and that the change of the care recipient was upsetting (57%). Moreover, answers on the positive CSI+ items revealed that most caregivers received appreciation from their loved one for providing care (84%). Further, the majority of caregivers found caring important (98%) and were happy to provide the care (91%). Many caregivers could handle the care fine (65%) and slightly more than half of the caregivers had enough time for themselves (52%).

Table 6.5 Bivariate association of Carer Experience Scale (CES) and CarerQol-7D overall score with care situation, caregiver and care recipient characteristics in Spearman's correlation coefficients or mean values, n ranges from 88 to 94

			CES	CarerQol-7D overall score
Care situation				
Relationship	partner		74.1	75.6
	parent		71.0	68.1
	other		68.0	79.9
Main caregiver	yes		72.2	74.0
	no		77.0	72.3
Sharing household	yes		72.7	73.8
	no		71.9	74.1
Years			-0.15	-0.30^{**}
Hours p/wk			0.01	0.06
Caregiving tasks	help with daily living	yes	71.6	73.3
		no	75.3	75.1
	housework	yes	72.1	73.2
		no	74.3	75.8
	organisational tasks	yes	71.9	73.2
		no	76.2	77.1
	other tasks	yes	73.1	72.7
		no	70.3	78.1
Motivation	duty	yes	74.0	72.5
		no	70.5	74.7
	closest person	yes	72.8	74.6
		no	70.5	67.7
	free choice	yes	73.2	75.7
		no	71.3	69.6
	no-one else	yes	71.6	71.2
		no	73.1	75.4
	no money paid care	yes	72.3	62.6
		no	72.5	76.5
Caregiver				
Age			0.02	0.18^{a}
Gender	female		71.9	71.6
	male		73.9	80.1
Educational level	16 years or less		72.6	73.9
	17 or 18 years		71.2	71.2
	19 years or over		72.6	73.9
Employment	yes		73.1	73.5
	no		71.8	73.3
EQ-5D			0.45^{***}	0.63^{***}
Palliative care patient				
Age			0.05	-0.14
Gender	female		70.3	73.3
	male		74.6	74.3
Physical health problem only	yes		75.1^{*}	78.1^{**}
	no		68.6	68.1

Note: ethnicity was excluded given small numbers in non-Australian/European groups.
* p<0.05, ** p<0.01, *** p<0.001, ^{a} p<0.10

Table 6.6 The Carer Experience Scale (CES), CarerQol instrument, Process Utility (PU), Expanded Caregiver Strain Index (CSI+) scores in percentages or mean (SD), n=97

Instrument	Statistic	Value
Carer Experience Scale	Mean score (SD)	72.5 (16.3)
Activities	Most / some/ few (%)	27 / 33 / 40
Social support	A lot / some / little (%)	46 / 38 / 15
Institutional support	A lot / some / little (%)	26 / 48 / 26
Fulfilment [b]	Mostly / sometimes / rarely (%)	63 / 28 / 7
Control [a]	Most / some / few aspects (%)	55 / 36 / 8
Getting on [b]	Mostly / sometimes / rarely (%)	87 / 10 / 1
CarerQol	Mean CarerQol-VAS score (SD)	5.8 (2.0)
	Mean CarerQol-7D overall score (SD)	73.5 (17.1)
Fulfilment [b]	No / some / a lot (%)	6 / 36 / 56
Relational problems [c]	No / some / a lot (%)	52 / 38 / 6
Mental health problems [a]	No / some / a lot (%)	29 / 57 / 13
Problems daily activities [b]	No / some / a lot (%)	25 / 56 / 18
Financial problems [a]	No / some / a lot (%)	61 / 25 / 13
Support [a]	No / some / a lot (%)	13 / 58 / 29
Physical health problems [a]	No / some / a lot (%)	29 / 55 / 15
Process Utility	Mean score (SD)	1.4 (4.5)
	Negative scores (%)	36
	Neutral scores (%)	6
	Positive scores (%)	56
	Missing scores (%)	2
Expanded Caregiver Strain Index [d]	Mean sum score negative items CSI+ (SD)	5.9 (3.4)
	Sum score negative items CSI+ >=7 (%)	44.3
	Mean sum score positive items CSI+ (SD)	4.0 (0.9)
Sleep is disturbed	Yes (%)	40
Inconvenient	Yes (%)	26
Recipient appreciates care	Yes (%)	84
Physical strain	Yes (%)	29
Confining	Yes (%)	63
Enough time for self	Yes (%)	52
Family adjustments	Yes (%)	48
Changes in personal plans	Yes (%)	72
Other demands on time	Yes (%)	46
Emotional adjustments	Yes (%)	35
Handle the care fine	Yes (%)	65
Behaviour upsetting	Yes (%)	38
Recipient change upsetting	Yes (%)	57
Happy to care	Yes (%)	91
Work adjustments	Yes (%)	37
Financial strain	Yes (%)	35
Feel completely overwhelmed	Yes (%)	44
Care is important	Yes (%)	98

Note: positive items of Expanded Caregiver Strain Index (CSI+) are in italics.
[a] 1% missing values, [b] 2% missing values, [c] 4% missing values, [d] range missing values 1% - 8%

6.4 Discussion

This chapter had two main aims: (i) investigating the construct validity of the CES and CarerQol in caregivers in end-of-life care for use as outcome measures in economic evaluations, and (ii) documenting the effect of providing informal care on caregivers of palliative care patients.

The construct validation results of these two preference-based caregiver outcome instruments for use in economic evaluations, confirm the hypotheses about correlates of the effect of caregiving in end-of-life care. That is, as a priori expected [41, 46-48], the CES, the CarerQol-7D overall score and the CarerQol-VAS were associated with other measures of caregiving burden: caregivers with better caring experiences generally had lower burden scores on the negative items of the CSI+ instrument, higher process utility of caring (PU), and more positive experiences from caring (positive items CSI+). Moreover, as hypothesized, convergent and discriminative validation tests showed that the CES and the CarerQol-7D overall score measured constructs that highly resembled each other.

Concerning convergent validity of the individual items of the CES, CarerQol-7D and the CSI+ items, almost all hypotheses were confirmed and the majority of associations of the single CES and CarerQol-7D dimensions that were not a priori expected had small strength of association. Nevertheless, only two of the a priori expected associations between the CES and the CarerQol-7D were (very) strongly associated, most correlation coefficients had moderate strength. One hypothesized association, between 'performing activities outside caring' and CSI+ item 'other demands on time', was only confirmed by the CarerQol-7D and not by the CES. Possibly, the constructs measured here differed. That is, the CSI+ item only registers whether caregivers perform other activities, regardless of the problems encountered in combination with caregiving.

This chapters shows that, as expected, caregivers in palliative care experienced a higher negative impact of caring when the patient suffered from mental health, intellectual health or multiple types of health problems than when they experienced only physical health problems. The results indicate that caregivers experienced a lower negative impact of caregiving when their own health was better. The influence of patients' and caregivers' health on caring experiences is also confirmed by other CES or CarerQol validation studies (chapters 3 and 4, [110, 111]). This study also shows that caregivers caring for longer periods of time experienced more negative caring experiences on the CarerQol-7D, as some previous CES or CarerQol validation studies also seem to indicate (chapter 4, [111]). Moreover, this study found that older caregivers seemed to have less negative impact of caring, measured on the CarerQol-7D. The hypothesized association between the effect of caring and the type of relationship between caregiver and care recipient was not confirmed. An explanation for this could be that most caregivers and patients were close relatives in the sample used in this study.

Given the results of this study and the specific application of the validation tests within this specific group of caregivers, this study cannot conclude which instrument performs better to measure the caring experience for inclusion in (economic) evaluation studies in palliative care. Both the CES and the CarerQol need further testing of psychometric properties and application in informal care research in different study settings.

A note should be made on the use of the CES and the CarerQol in economic evaluations of health care programs. In economic evaluations effects of patients typically are measured in terms of health effects with preference based instruments of health such as the EQ-5D [70]. Although informative on health effects, this instrument does not include important caregiver aspects such as relationships and fulfilment and may be insensitive to the psychological effects and positive impacts of caregiving [113]. The CES and the CarerQol record the full impact of informal care. Moreover, both instruments measure a preference based outcome specifically for caregivers. Although the CES and CarerQol value caregiver outcomes in economic terms, CES and CarerQol scores cannot straightforwardly be added to health effects in patients in economic evaluations in general. Nevertheless, if patient's utility is central in interventions, the CES or the CarerQol can be used to include caregiver effects in economic evaluations using multi-criteria analysis (MCA). In MCA different outcomes, including effects in patients and their caregivers, can be used to evaluate palliative care programmes (chapter 8, [25, 29, 85]). Moreover, the CES or CarerQol can be included as an outcome in cost-effectiveness or cost-utility analysis studies if the caregiver's utility is the main objective of the intervention.

Concerning the second research question, this study shows that caregivers in palliative care often felt strained by caregiving. Over 40 per cent of caregivers experienced substantial strain (sum score negative items CSI+ >= 7). Caregivers often found it difficult to combine caregiving with other activities of daily life, such as work. Furthermore, many caregivers suffered from health problems themselves. The literature supports these findings [215, 216]. Regardless of this relatively high level of strain of caregiving in palliative care, many caregivers preferred to continue lending care. Caregivers in this study often received support from others with this care. Nevertheless, this study provides some evidence that not all caregivers seem to receive support when they need it. From this study, no information on the kind of additional support carers required is available, but other Australian studies found that caregivers in palliative care often have a need for respite care or more information on what to expect in the care process [215, 242].

This study has a number of limitations. First, the sample size was relatively small and this has implications for the generalizability of the findings. Caregivers in this study predominantly were older females taking care of their partner or parent with a physical health problem. Further research should be conducted in larger and more diverse end-of-life caregiving samples to verify the findings of this study. Secondly, although this chapters reports information on the informal care situation, no information is available on the type of other care consumed by the care recipient either at the time of questionnaire completion or previously, e.g., hospital or community services.

In conclusion, this study showed that both the CES and the CarerQol seem to capture the effect of caregiving in end-of-life care in a valid manner for inclusion in economic evaluations, without a clear indication that one instrument performs better than the other. To substantiate these findings, it is important that further validation research is conducted in larger and more varied samples of palliative caregivers. Furthermore, especially in the context of palliative care where family and friends are confronted with many physical and mental challenges given their loved ones' suffering and nearing end of life, the influence of these challenges on caregivers should be studied in more depth. For example, further research could separate this effect into two components: (i) the impact of lending care (so-called caregiving effect), and (ii) the effect of the bad health status of the patient (family effect) [50, 53]. Important to note is that the family effect is not restricted to caregivers, but could also occur in other family members and friends of patients [50, 53, 87, 88]. Valid measurement of the effect of caregiving on caregivers is important as it facilitates research that will improve our understanding of the needs of end-of-life caregivers and, hence, better support them. Moreover, it enables comparative (cost)-effectiveness research of palliative care interventions including both effects in patients and their caregivers. This is essential information for stimulating welfare enhancing policy decisions, especially in end-of-life care with patients receiving intensive care and support from their family and friends [225, 243].

Acknowledgements

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Appendix chapter 6

PLEASE TICK ONE BOX FOR EACH GROUP to indicate which statement best describes your current caring situation.

1.Activities outside caring (Socialising, physical activity and spending time on hobbies, leisure or study)

You can do most of the other things you want to do outside caring ☐ 1
You can do some of the other things you want to do outside caring ☐ 2
You can do few of the other things you want to do outside caring ☐ 3

2. Support from family and friends (Personal help in caring and/or emotional support from family, friends, neighbours or work colleagues)

You get a lot of support from family and friends ☐ 1
You get some support from family and friends ☐ 2
You get little support from family and friends ☐ 3

3. Assistance from organisations and the Government (Help from public, private or voluntary groups in terms of benefits, respite and practical information)

You get a lot of assistance from organisations and the Government ☐ 1
You get some assistance from organisations and the Government ☐ 2
You get little assistance from organisations and the Government ☐ 3

4. Fulfilment from caring (Positive feelings from providing care, which may come from: making the person you care for happy, maintaining their dignity, being appreciated, fulfilling your responsibility, gaining new skills or contributing to the care of the person you look after)

You mostly find caring fulfilling ☐ 1
You sometimes find caring fulfilling ☐ 2
You rarely find caring fulfilling ☐ 3

5. Control over the caring (Your ability to influence the overall care of the person you look after)

You are in control of most aspects of the caring ☐ 1
You are in control of some aspects of the caring ☐ 2
You are in control of few aspects of the caring ☐ 3

6. Getting on with the person you care for (Being able to talk with the person you look after, and discuss things without arguing)

You mostly get on with the person you care for ☐ 1
You sometimes get on with the person you care for ☐ 2
You rarely get on with the person you care for ☐ 3

Figure A6.1 Carer Experience Scale (CES)

We would like to form an impression of your caregiving situation.
Please tick a box to indicate which description best fits your caregiving situation at the moment.

Please tick only one box per description: 'no', 'some' or 'a lot of'.

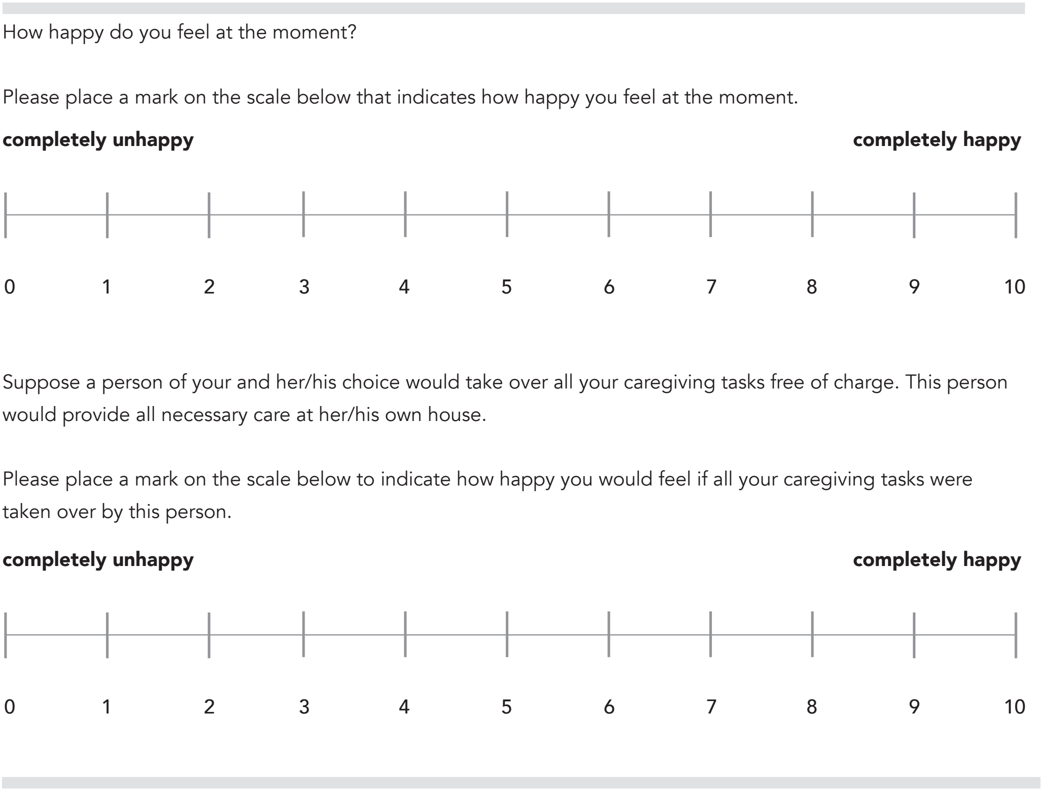
	no	**some**	**a lot of**	
I have	☐	☐	☐	fulfilment from carrying out my care tasks.
I have	☐	☐	☐	relational problems with the care receiver (*e.g., he/she is very demanding or he/she behaves differently; we have communication problems*).
I have	☐	☐	☐	problems with my own mental health (*e.g., stress, fear, gloominess, depression, concern about the future*).
I have	☐	☐	☐	problems combining my care tasks with my daily activities (*e.g., household activities, work, study, family and leisure activities*).
I have	☐	☐	☐	financial problems because of my care tasks.
I have	☐	☐	☐	support with carrying out my care tasks, when I need it (*e.g., from family, friends, neighbours, acquaintances*).
I have	☐	☐	☐	problems with my own physical health (*e.g., more often sick, tiredness, physical stress*).

How happy do you feel at the moment?

Please place a mark on the scale below that indicates how happy you feel at the moment.

completely unhappy **completely happy**

Figure A6.2 The CarerQol instrument



How happy do you feel at the moment?

Please place a mark on the scale below that indicates how happy you feel at the moment.

completely unhappy **completely happy**

0 1 2 3 4 5 6 7 8 9 10

Suppose a person of your and her/his choice would take over all your caregiving tasks free of charge. This person would provide all necessary care at her/his own house.

Please place a mark on the scale below to indicate how happy you would feel if all your caregiving tasks were taken over by this person.

completely unhappy **completely happy**

0 1 2 3 4 5 6 7 8 9 10

Figure A6.3 Process Utility (PU)

Below we present a list of statements from other people providing informal care. We are interested in how you feel about these statements. Please take the last week as reference. There are no right or wrong answers; we are interested in your view.

Sleep is disturbed	☐	No	☐	Yes
It is inconvenient	☐	No	☐	Yes
She/he appreciates everything I do for her/him	☐	No	☐	Yes
It is a physical strain	☐	No	☐	Yes
It is confining	☐	No	☐	Yes
Besides the care I provide to her/him, I have enough time for myself	☐	No	☐	Yes
There have been family adjustments	☐	No	☐	Yes
There have been changes in personal plans	☐	No	☐	Yes
There have been other demands on my time	☐	No	☐	Yes
There have been emotional adjustments	☐	No	☐	Yes
I can handle the care for her/him fine	☐	No	☐	Yes
Some behaviour is upsetting	☐	No	☐	Yes
It is upsetting to find that she/he has changed so much from her/his former self	☐	No	☐	Yes
I am happy to care for her/him	☐	No	☐	Yes
There have been work adjustments	☐	No	☐	Yes
Feeling completely overwhelmed	☐	No	☐	Yes
It is a financial strain	☐	No	☐	Yes
Taking care for her/him is important to me	☐	No	☐	Yes

Figure A6.4 Expanded Caregiver Strain Index (CSI+)

CHAPTER

7

A Discrete Choice Experiment to obtain a tariff for valuing informal care situations measured with the CarerQol instrument

Based on Hoefman, R. J., van Exel, J., Rose, J. M., van de Wetering, E. J., & Brouwer, W. B. (2014). A Discrete Choice Experiment to Obtain a Tariff for Valuing Informal Care Situations Measured with the CarerQol Instrument. Medical Decision Making, 34(1), 84-96.

Abstract

Background/Objective

Economic evaluations adopting a societal perspective need to include informal care, whenever relevant. However, in practice, informal care is often neglected, because there are few validated instruments to measure and value informal care for inclusion in economic evaluations. The CarerQol, which is such an instrument, which measures the impact of informal care on seven important burden dimensions (CarerQol-7D) and values this in terms of general quality of life (CarerQol-VAS). The objective of this study was to calculate utility scores based on relative utility weights for the CarerQol-7D. These tariffs will facilitate inclusion of informal care in economic evaluations.

Methods

The CarerQol-7D tariff was derived with a Discrete Choice Experiment (DCE) conducted as an Internet survey among the general adult population in the Netherlands (n=992). The choice set contained two unlabelled alternatives described in terms of the seven CarerQol-7D dimensions (level range: 'no', 'some' and 'a lot'). An efficient experimental design with priors obtained from a pilot study (n=104) was used. Data was analysed with a panel mixed multinomial parameter model including main and interaction effects of the attributes.

Results

The utility attached to informal care situations was significantly higher when this situation was more attractive in terms of fewer problems and more fulfilment or support. The interaction term between the CarerQol-7D dimensions physical health and mental health problems also significantly explained this utility. The tariff was constructed by adding up the relative utility weights per category of all CarerQol-7D dimensions and the interaction-term.

Conclusions

We obtained a tariff providing standard utility scores for caring situations described with the CarerQol-7D. This facilitates the inclusion of informal care in economic evaluations.

7.1 Introduction

Informal care provision can significantly affect the lives of caregivers, both positively and negatively [14, 28, 42]. Hence, economic evaluations adopting a societal perspective need to include the impact of informal care in order to ensure a full assessment of costs and benefits related to health care interventions. However, despite the increased attention on informal care in economic evaluations [74, 75, 77, 78], it still remains frequently omitted, and it is also omitted in evaluations adopting a societal perspective [92]. Obviously, ignoring informal care in economic evaluations may subsequently result in nonoptimal decisions, potentially at the expense of informal caregivers.

One of the explanations for omitting informal care from economic evaluations may be that most outcome measures used in these evaluations are either difficult to implement or ill-suited to capture the full effects of interventions on informal caregivers [91, 96, 97]. Frequently used methods to value informal care in monetary terms, such as the opportunity cost method and the shadow price method, do not reflect the full impact of informal care or caregiver preferences [91, 99]. Other monetary valuation methods, such as the contingent valuation method [116, 117], conjoint analysis with a monetary component [118] or the well-being method [119], all with their own methodological challenges, allow the inclusion of a more preference-based impact of informal care in monetary terms in cost-benefit analysis (requiring all impacts to be expressed in monetary terms) or on the cost side of more common cost-effectiveness analyses. However, these methods commonly provide little direct information regarding the underlying problems that informal caregivers experience (e.g., lack of support, financial problems or relational problems) [25, 26]. Moreover, given the inclusion of these methods on the cost side of the cost-effectiveness-ratio (CE-ratio), as one of the normally numerous cost items relative to normally few non-monetary outcomes, applying these methods may also be perceived to result in relatively little explicit attention for informal care.

Alternatively, non-monetary methods can be used to describe the impact of caregiving in more detail. In that case, the impact (full or partial) of informal care is captured in terms of effects rather than costs and is placed on the effect side of the CE-ratio. Such shifts are possible (and have been debated in other contexts as well, e.g., Brouwer et al. [244]), as long as all effects are soundly captured and double counting is avoided. In the context of informal care, it is possible, for instance, to measure health changes in caregivers in terms of quality-adjusted life-years (QALYs), using a generic health-related quality of life instrument like the EuroQol-5D instrument [122]. These could subsequently be included in cost-utility analysis (combined with or separate from QALY changes in patients). However, this results in a partial valuation of informal care, since QALYs only capture the health effect, whereas caring influences more aspects of life than health alone [25, 26]. Hence, subjective burden instruments that aim to capture all important aspects of burden, such as social and financial problems, are often used in the context of informal care [26, 104, 105]. While these measures aim to provide a rich description of the effect of informal care, most instruments only focus on negative aspects of caregiving and not on positive ones. In addition, most measures result in some unweighted sum score and do not provide an economic valuation of the impact of caregiving.

Hence, there appeared to be a need for instruments combining the informational density of subjective burden measures with an economic valuation component. Only two such instruments have been developed to date to our knowledge: the Carer Experience Scale (CES) [108] and the Care-related Quality of Life instrument (CarerQol) [110]. The CarerQol instrument combines a description of the caregiving situation (CarerQol-7D) with a valuation component in terms of general well-being (CarerQol-VAS) [110]. The CarerQol instrument is shown in Figure 7.1. The development of the CarerQol was based on that of the EuroQol instrument [122]. The CarerQol-7D measures subjective burden on seven dimensions (+ or - indicating positive or negative dimension): fulfilment (+), relational problems (-), mental health problems (-), problems with combining daily activities (-), financial problems (-), social support (+) and physical health problems (-) of caregiving. For each of these dimensions three response categories are defined ('no', 'some', 'a lot'), so that a total of 2,187 (= 3^7) different 'informal care situations' can be distinguished. The CarerQol-VAS is a Visual Analogue Scale (VAS) measuring general quality of life in terms of happiness, ranging from 0 ('completely unhappy') to 10 ('completely happy') and with numeric anchors equally spaced between these two extreme levels. The main advantage of such a broad outcome measure is that the different effects of caregiving, such as health or financial problems, can all be incorporated and caregivers can indicate the influence of informal care on happiness. This chapter does not focus on the VAS scale but aims at deriving standard utility scores for the care states described with the CarerQol-7D. To date, the CarerQol has been included in several studies [54, 163-167] and its psychometric properties were shown to be satisfactory (chapters 2 and 3, [110]). Standard utility scores for the care situations described with the CarerQol would facilitate and improve its use for several reasons [110]. First, responses to the broad outcome measure of happiness (CarerQol-VAS) are also influenced by effects outside the direct scope of caregiving, such as income level, type of job and social contacts. Therefore, utility scores exclusively based on the caregiving domains of the CarerQol-7D would directly reflect purely 'informal care situation-related utility'. Secondly, like patients, informal caregivers may adapt to and cope with their care situation, which would result in upwardly biased happiness scores. Tariffs obtained in the general public would be less influenced by coping and adaptation, hence providing a more standardized reflection of informal care situation valuations. Note that important discussions are ongoing in the literature regarding whose preferences to use in deriving health state valuations: those of the group affected (e.g., patients or informal caregivers) or those of the general public [70]. We collected the values of the general population to resemble the current practice of health utilities elicitation, and in specific the method of the EuroQol-instrument (e.g., [130, 131]).

To date, no tariff for the CarerQol exists. For the CES, index values were obtained for its descriptive system using preferences of caregivers of older people in the United Kingdom collected with a discrete choice experiment (DCE) [113]. The aim of this study is to derive a tariff for the CarerQol-7D, using a DCE in a large and representative sample of the Dutch general public. In a DCE, respondents are asked to choose between hypothetical options [245], thus reflecting their preferences. The tariff elicited in this study will enable researchers to calculate utility scores for CarerQol-7D, taking into account differences in severity of problems, and will therefore facilitate the inclusion of informal care in economic evaluations of health care interventions.

We would like to form an impression of your caregiving situation.
Please tick a box to indicate which description best fits your caregiving situation at the moment.

Please tick only one box per description: 'no', 'some' or 'a lot of'.

	no	**some**	**a lot of**	
I have	☐	☐	☐	fulfilment from carrying out my care tasks.
I have	☐	☐	☐	relational problems with the care receiver (*e.g., he/she is very demanding or he/she behaves differently; we have communication problems*).
I have	☐	☐	☐	problems with my own mental health (*e.g., stress, fear, gloominess, depression, concern about the future*).
I have	☐	☐	☐	problems combining my care tasks with my daily activities (*e.g., household activities, work, study, family and leisure activities*).
I have	☐	☐	☐	financial problems because of my care tasks.
I have	☐	☐	☐	support with carrying out my care tasks, when I need it (*e.g., from family, friends, neighbours, acquaintances*).
I have	☐	☐	☐	problems with my own physical health (*e.g., more often sick, tiredness, physical stress*).

How happy do you feel at the moment?

Please place a mark on the scale below that indicates how happy you feel at the moment.

Figure 7.1 The CarerQol instrument

7.2 Methods

7.2.1 Discrete choice experiment

DCEs are based on the random utility model developed by McFadden [246], assuming that respondents compare different alternatives and choose the one with the highest latent utility value [70, 247]. The latent utility value of the jth-respondent is formulated as:

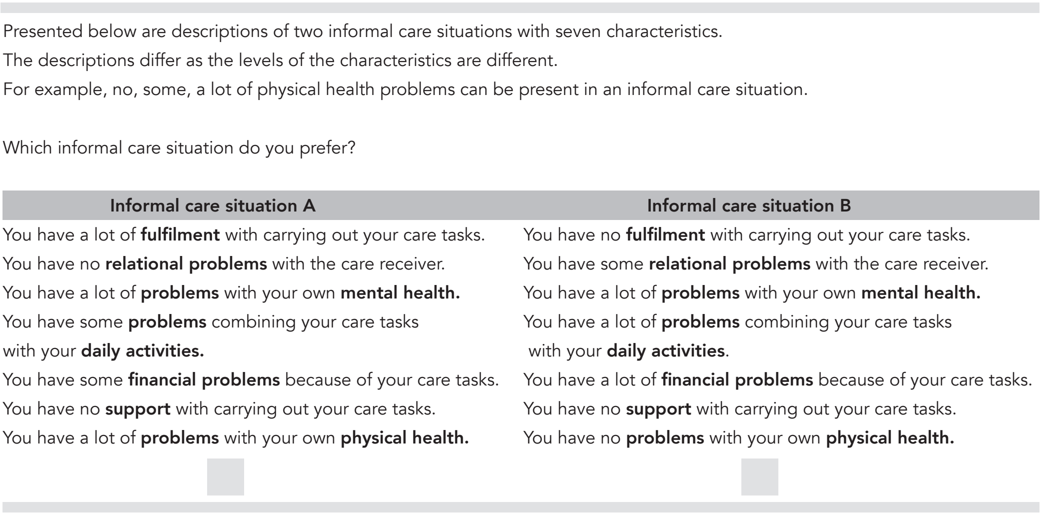
$$Uj = \lambda * Vj + \varepsilon j$$

with Vj as the systematic component reflecting observed influences of attributes and levels, εj as the stochastic component reflecting unobserved influences, and λ as the scale parameter reflecting the variance of the unobserved influences [247].

7.2.1.1 Choice task

In this study, the choice tasks consisted of paired comparisons of two unlabelled descriptions of hypothetical informal care situations (see Figure 7.2 for an example). The attributes and levels were derived from the CarerQol-7D: (i) fulfilment with carrying out your care tasks, (ii) relational problems with the care receiver, (iii) problems with your own mental health, (iv) problems combining your care tasks with your daily activities, (v) financial problems because of your care tasks, (vi) support with carrying out your care tasks, and (vii) problems with your own physical health. The attribute level range was: 'no', 'some' and 'a lot' for all attributes. For clearer graphical presentation, colours were added to these levels: green for attribute levels regarded positive (i.e., 'a lot' for the positive and 'no' for the negative attributes), red for those regarded negative (i.e., 'no' for the positive and 'a lot' for the negative attributes), and orange for the intermediate level 'some'.

Prior to the choice tasks, respondents received information on what informal care was, which care activities were commonly performed, time typically spent on informal care, and the fulfilment and burden derived from caregiving in the Netherlands. In addition, the attributes were explained with some examples. Next, respondents were instructed to imagine that they were informal caregivers and that they provided care to the same person during the whole experiment. The complete text of these instructions was shown to all respondents for at least five seconds before they could continue. To reduce the effect of order-biases on the results, the order in which the choice sets were presented to respondents was randomized.



Presented below are descriptions of two informal care situations with seven characteristics.
The descriptions differ as the levels of the characteristics are different.
For example, no, some, a lot of physical health problems can be present in an informal care situation.

Which informal care situation do you prefer?

Informal care situation A	Informal care situation B
You have a lot of **fulfilment** with carrying out your care tasks.	You have no **fulfilment** with carrying out your care tasks.
You have no **relational problems** with the care receiver.	You have some **relational problems** with the care receiver.
You have a lot of **problems** with your own **mental health.**	You have a lot of **problems** with your own **mental health.**
You have some **problems** combining your care tasks with your **daily activities.**	You have a lot of **problems** combining your care tasks with your **daily activities**.
You have some **financial problems** because of your care tasks.	You have a lot of **financial problems** because of your care tasks.
You have no **support** with carrying out your care tasks.	You have no **support** with carrying out your care tasks.
You have a lot of **problems** with your own **physical health.**	You have no **problems** with your own **physical health.**
☐	☐

Figure 7.2 Example of a choice set.

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[3] We used orthogonal array 27.13.3.2. We assigned the first seven columns of the orthogonal array to the seven attributes (CarerQol-7D), used one column for the blocking strategy and deleted the remaining five columns of the orthogonal array. The orthogonal array provided only the order of the first alternative in the choice set. The second alternative was constructed by a fold-over (0=1, 1=2, 2=0) of the orthogonal array.

7.2.1.2 Questionnaire

Our questionnaire consisted of different parts, of which the DCE was the first and main part. After the actual experiment, respondents were asked whether they knew what informal care was prior to participating in this research and whether they were an informal caregiver or care recipient themselves. Next, respondents were asked whether they as hypothetical caregivers had (i) a specific person in mind as the care recipient and, if so, (ii) whom and whether they (iii) shared a household with this person. And whether they considered (iv) the number of years caregiving, (v) number of hours per week caring, and (vi) the type of care activity they would perform as a hypothetical caregiver. Background characteristics of respondents were age, gender, highest attained educational level, yearly household income, partner status, having children, performing paid and unpaid work, having a private household help, health status measured with the EQ-5D [122] and general well-being in terms of happiness, measured with the CarerQol-VAS [110] (Table 7.1).

7.2.2 Pilot study

In March 2011, data was gathered from 104 respondents with the Web-based questionnaire distributed by an Internet survey company in a representative sample of adult persons in the Netherlands in terms of age and gender. The aim of the pilot was twofold: (i) collecting prior information for an efficient design for the main study [248] and (ii) pretesting the questionnaire and gaining an indication of its feasibility and the quality of the data.

The choice tasks were constructed using an orthogonal main effects plan (OMEP)[3] with 27 questions. A blocking strategy was used to distribute levels among alternatives, and provided nine choice sets among three respondent groups. A fractional factorial design was chosen, because gathering data on all 2,187 alternatives is practically impossible.

The random utility model was operationalized with conditional logit regression (also called fixed effects logit or multinomial logit (MNL)) with the error term assumed to be identically independently distributed (IID) with an extreme value type 1 distribution (EV1)[4] in Stata® version 11.0 (StataCorp LP). In this model, scale is normalized to 1, implying constant error variance. Maximum likelihood estimations were calculated for the dependent variable (binary variable with 1 'alternative is chosen by respondent *n* in choice set *s*' or 0 'otherwise') per choice set.

7.2.3 Main study

7.2.3.1 Data

The data of the main study was collected in June 2011 with a Web-based questionnaire distributed by an Internet survey company to a representative sample, in terms of age and gender, of adult persons in the Netherlands. In total, 992 respondents[5] were included in the data set.

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[4] We used the clogit command with the group option in Stata.

[5] We excluded 23 respondents from the final data set because of concerns regarding the quality of the data given extremely short completion time (the minimum participation time to be included in the data set was 10 minutes for the full questionnaire).

Table 7.1 Background characteristics study sample, n=992

		n (%)	mean (SD)
Age			49.2 (16.0)
Gender	female	596 (60.1)	
	male	396 (39.9)	
Educational level	primary/no	189 (19.1)	
	lower/medium vocational	523 (52.7)	
	higher vocational/university	280 (28.2)	
Income	low	395 (39.8)	
	middle	258 (26.0)	
	high	70 (7.1)	
	missing	269 (27.1)	
Partner	yes	676 (68.2)	
	no	316 (31.9)	
Children in household	yes	289 (29.3)	
	no	698 (70.7)	
Private household help	yes	106 (10.7)	
	no	886 (89.3)	
Paid work	full-time	198 (20.0)	
	part-time	255 (25.7)	
	no	539 (54.3)	
Unpaid work	yes	306 (30.9)	
	no	686 (69.2)	
Subjective health (0-10)			7.1 (1.6)
Health (EQ-5D)	no problems with mobility	732 (73.8)	
	problems with mobility	260 (26.2)	
	no problems with self-care	938 (94.6)	
	problems with self-care	54 (5.4)	
	no problems with usual activities	738 (74.4)	
	problems with usual activities	254 (25.6)	
	no experience of pain/discomfort	499 (50.3)	
	experience of pain/discomfort	493 (49.7)	
	no feelings of anxiety/depression	790 (79.6)	
	feelings of anxiety/depression	202 (20.4)	
Happiness (0-10)			7.4 (1.5)

7.2.3.2 Design

An efficient experimental design with priors from the MNL model of the pilot study for all attributes was used (Table 7.2) to construct the choice sets and to calculate the standard errors of the parameters as statistically efficiently as possible [249], which generally improves the reliability of the results with smaller sample sizes [248-250]. The efficient design was created in Ngene® (ChoiceMetrics 2011). The 40 choice sets[6] were randomly distributed over four respondent groups; thus, there were ten per group. The specified utility functions for the two alternatives included fourteen dummy variables for the seven attributes, 21 interaction terms for all attribute combinations, and a constant term for the first alternative. The attributes were dummy coded to allow construction of a utility score for the CarerQol. The attributes with the highest standard errors (see Table 7.2) were treated as Bayesian priors: relational problems ('no'), mental health problems ('some'), problems with daily activities ('no'), support ('a lot'),

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[6] We used the decision rule of S x (J-1) ≥ K, where S=number of choice sets, J=number of alternatives and K=degrees of freedom of parameters, for choosing the minimum number of independent choice probabilities.

and physical health problems ('no', 'some')[7]. We used a normal distribution form for the Bayesian parameters. Mean values were calculated and 500 Halton draws were used to generate the design [251]. The design was optimized for D-efficiency in the basic multinomial logit model [249].

7.2.4 Analyses

We used a panel mixed multinomial parameter model (MMNL) to operationalize the random utility model. This model is less restrictive than the MNL model, allows for presence of unobservable preference heterogeneity in the sampled population by using random parameters (RPs) and considers the possibility of correlated responses across observations by the panel structure of the model [247, 252]. We tested several utility specifications of the panel MMNL with likelihood ratio (LR) tests. First, an alternative specific constant was included. Although it has been argued that this strategy violates the meaning of unlabelled experiments [247], it has been used to test for biases in an experiment, such as left-right biases and a biased experimental design [253]. In the model of this study, the constant was not significant and therefore was excluded. Secondly, all parameters were assumed to be random. Therefore, scale is also assumed to be random in the model, because scale is perfectly confounded with RPs [245, 254]. Moreover, the RPs indicated preference heterogeneity [252]. Consequently, in an attempt to find a potential source of this variability, interaction terms were added to the model of this study [252]. Including all interaction terms of the fourteen dummy variables led to estimation problems. Hence, the 21 combinations of all attributes considered as continuous variables, for example, fulfilment * mental health problems, were first included. The combinations of attributes of significant interaction terms were subsequently studied in more detail by including interaction terms per level combination of these attributes, for example, some fulfilment * no relational problems. Nonsignificant interaction terms were excluded from the model. Thirdly, an LR test examining an N3-term, that is, a correction for having a worst level in at least one of the dimensions, as is common in the utility specification for the EQ-5D [130, 131], showed that such an N3-term was not statistically significant here. Fourth, it was tested whether attribute levels could be collapsed. The utility specification used in this study was as follows:

$$U = \beta 1 fulfilment(some) + \beta 2 fulfilment(a\ lot) + \beta 3 relational(no) + \beta 4 relational(some) + \beta 5 mental(no) + \beta 6 mental(some) + \beta 7 daily(no) + \beta 8 daily(some) + \beta 9 financial(no) + \beta 10 financial(some) + \beta 11 support(some) + \beta 12 support(a\ lot) + \beta 13 physical(no/some) + \beta 14 mental(no)*physical(no)$$

7.2.5 CarerQol tariff

The parameters of the panel MMNL model represent population level estimates (unconditional distribution). To calculate the tariff, the random parameter distribution was reconstructed out of the study sample [247]. This unconditional distribution was randomly assigned over a hypothetical sample of 10,000 individuals with bootstrap sampling. The resulting individual-specific parameter estimates were averaged and rescaled to represent the CarerQol-7D tariff. This was done by: (i) adding up the mean beta coefficients of the best informal care situation (i.e., 'a lot' for fulfilment and support and 'no' for the five problem dimensions) and the interaction term; (ii) dividing all estimates by this total score. Next, these relative scores were transformed to represent a utility score between 0 (i.e., worst informal care situation as defined by the CarerQol-7D) and 100 (i.e., best informal care situation). The standard errors of the tariff were calculated by dividing the standard errors of the MMNL parameters by the same total score. Analyses were performed in Nlogit® version 5.0 (Econometric Software Inc.).

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[7] We used mean parameter estimates of the MNL model for the priors (Table 2). For Bayesian priors we used both the mean and standard error of the MNL model of the pilot study.

Table 7.2 Results of conditional logistic model of choice of informal care situation based on the CarerQol-7D, n=104

		coef.	se	p-value	95% c.i.	
Fulfilment [a]	some	0.35	0.11	0.002	0.13	0.58
	a lot	0.89	0.11	0.000	0.67	1.12
Relational problems [b]	no	0.54	0.12	0.000	0.31	0.78
	some	0.50	0.12	0.000	0.28	0.73
Mental health problems [b]	no	0.84	0.12	0.000	0.61	1.06
	some	0.61	0.12	0.000	0.38	0.84
Problems daily activities [b]	no	0.33	0.12	0.006	0.10	0.56
	some	0.29	0.11	0.011	0.07	0.51
Financial problems [b]	no	0.85	0.12	0.000	0.62	1.08
	some	0.61	0.12	0.000	0.38	0.84
Support [a]	some	0.40	0.11	0.000	0.18	0.62
	a lot	0.62	0.12	0.000	0.38	0.86
Physical health problems [b]	no	0.82	0.12	0.000	0.59	1.05
	some	0.81	0.12	0.000	0.57	1.05
pseudo R^2		0.25				

Note: The conditional logit model does not report a constant, because the constant is stable among the alternatives within a choice set per respondent. Consequently, the constant is cancelled out in the utility specification model.
[a] reference category: 'no'; [b] reference category: 'a lot'

7.3 Results

7.3.1 Study sample

Background characteristics of the sample of this study are presented in Table 7.1. The mean age of respondents included in this study was about 49 years, and 60% were female. Most of the respondents had a medium or high educational level. Many respondents had a relatively low income, although this figure might be biased, because a large proportion of respondents (27%) did not know or did not want to reveal this.

7.3.2 Context

Information on the context respondents had in mind during the experiment is presented in Table 7.3. While imagining that they would provide informal care, more than half of the respondents thought of a person in their social network, often their parent (38%) or partner (26%), to whom they would provide this care. Thirty percent of the respondents shared a household with this person. Many respondents imagined they would perform specific care activities. The majority of the study sample did not make an assumption regarding the intensity or duration of caregiving.

7.3.3 Concept of informal care

Almost all respondents knew what informal care was before they participated in the experiment (Table 7.3). Almost 16% were currently informal caregivers themselves.

Table 7.3 Description of the context of the choice experiment, n=992

		n	%
Specific person in mind		588	59.3
Specific person is:	partner	153	26.0
	parent	229	39.0
	parent-in-law	35	6.0
	child	29	4.9
	brother/sister	28	4.8
	grandparent	17	2.9
	uncle/aunt	10	1.7
	other family member	17	2.9
	neighbour	22	3.7
	friend/acquaintance	48	8.2
Shares household with specific person		181	30.8
Time spent caring in mind	hours (median)	350	35.3 (14)
	years (median)	181	18.6 (5)
Caregiving tasks in mind		603	61.0
Which task(s)? [a]	household activities	404	67.0
	personal care	377	62.5
	mobility	296	49.1
	administrative tasks	315	52.2
	social support	359	59.5
Familiarity of informal care			
Knowledge of concept of informal care		936	94.4
Role	not a caregiver or care recipient	388	39.1
	care recipient	29	2.9
	care recipient in past	16	1.6
	caregiver	158	15.9
	caregiver in distant past	43	4.3
	caregiver in past	124	12.5
	care recipient or caregiver in social network	234	23.6

[a] Adds up to more than 100%, because respondents could indicate more than one activity in the questionnaire

7.3.4 Relative weights of dimensions CarerQol-7D

The relative weights of the CarerQol-7D are presented in Table 7.4. In general, the utility of informal care situations is significantly higher when this situation is more attractive. The informal care situation-related utility is significantly higher when caregivers derive fulfilment from caregiving or receive support from others with their caregiving tasks. The utility of informal care situations is significantly higher when no or only some problems are present, compared with situations with a lot of problems. One interaction-effect was significant in the model: When both mental health and physical health problems are absent the informal care situation-related utility is significantly higher. The CarerQol-7D dimensions fulfilment, physical health problems, relational, and financial problems relatively had the strongest influence on informal care situation-related utility. Support had the weakest influence in that respect.Overall, the model correctly predicts 67% of the choices made (not presented in table).

Table 7.4 Coefficients of panel MMNL model, n=992

		MMNL unconditional parameter distributions		
		coef.	**sd [a]**	**p-value**
Fulfilment [b]	some	1.76	0.95	0.000
	a lot	2.55	1.72	0.000
Relational problems [c]	no	1.89	1.51	0.000
	some	1.38	1.24	0.000
Mental health problems [c]	no	1.73	1.36	0.000
	some	1.2	1.16	0.000
Problems daily activities [c]	no	1.29	1.09	0.000
	some	0.84	0.79	0.000
Financial problems [c]	no	1.86	1.77	0.000
	some	1.37	1.27	0.000
Support [b]	some	0.61	0.91	0.000
	a lot	0.86	1.09	0.000
Physical health problems [c]	no/some	1.94	1.66	0.000
Interaction term: no mental health problems * no physical health problems		0.86	1.78	0.000

Note: Each respondent answered 10 choice sets making a total number of observations of 9920 in analysis of the MMNL model.
[a] Attribute specific standard deviation of Cholesky matrix; [b] Reference category: 'no';
[c] Reference category: 'a lot'

Table 7.5 CarerQol tariff and standard errors per CarerQol-7D dimension

Dimension	**no**		**some**		**a lot**	
	tariff	se [a]	tariff	se [a]	tariff	se [a]
Fulfilment	0.0	0.0	13.6	1.1	19.7	1.4
Relational problems	14.7	1.4	10.6	1.0	0.0	0.0
Mental health problems	13.3	1.3	9.3	1.0	0.0	0.0
Problems daily activities	10	0.9	6.4	0.8	0.0	0.0
Financial problems	14.3	1.2	10.6	1.0	0.0	0.0
Support	0.0	0.0	4.7	0.8	6.5	0.9
Physical health problems	15.1	1.1	15.1	1.1	0.0	0.0
plus: interaction term	**no**		**yes**			
	tariff	se	tariff	se		
No mental health problems and no physical health problems	0.0	0.0	6.6	1.7		

[a] Rescaled standard errors of MMNL parameters

7.3.5 CarerQol tariff

The utility score of an informal care situation described by the CarerQol-7D can be calculated by starting with score 0 and adding up the tariff per category of all CarerQol-7D dimensions presented in Table 7.5. To derive the total score, a bonus score should be added if an informal care situation is characterized by the absence of both mental and physical health problems. For example, the utility score of an informal care situation in which the caregiver has 'some fulfilment, a lot of relational problems, no mental health problems, some problems combining daily activities, no financial problems, a lot of support and no physical health problems' is 75.7 (Table 7.5).

7.4 Discussion

In this study, relative utility weights for the CarerQol-7D were calculated to construct a tariff for this instrument. A carefully designed and pilot-tested discrete choice experiment was used to derive this information among the general public in the Netherlands. The utility derived from an informal care situation increases with higher scores on the two positive dimensions and decreases with more problems on the five negative dimensions. Important dimensions of utility were fulfilment, physical health problems, relational problems, and financial problems related to informal care. Moreover, physical health was even more important in combination with problems with mental health. Overall, the model performed well as two-thirds of the choices were correctly predicted.

To our knowledge, five studies applied conjoint analysis to value aspects of informal care to date. One study elicited index values for the CES [113], while four other studies derived monetary valuations of informal care with a ranking task [118, 255, 256] and a DCE [257]. Given the similarities in aim, it is interesting to compare the results of this study with those of Al-Janabi et al. [113]. Although such direct comparisons are difficult, it appears that some differences exist between the results of this study and those reported by Al-Janabi et al. (2011), in terms of the relative importance of the different subjective burden dimensions. Although performing daily activities and receiving support are important dimensions of informal care situation-related utility in their study, these dimensions had a relatively small impact in this study. Whether these differences indicate preference heterogeneity in preferences among countries (UK [113] versus the Netherlands) or type of respondents (caregivers [113] versus general public) remains unclear, because these results could also be caused by differences in the methods used. For example, this study included attributes on health, while the study of Al-Janabi et al. (2011) included an attribute on the level of control over the care situation. More research in this area remains warranted, therefore.

7.4.1 Study limitations

Before the results and implications of this study will be discussed in more detail, it is important to note some limitations of this study. First, despite using a representative study population, the study sample is not perfectly representative of the general population in the Netherlands, because females were somewhat overrepresented in the sample used in this study. The influence of this bias is likely to be minimal, given that analysing a subsample of the data with the same gender characteristics as the Dutch population showed highly comparable results. Secondly, attention should be paid to the issue of complexity of the choice tasks for respondents. Choice complexity in general, and answering multiple choice questions in particular, seem to distort parameter estimates by increasing error variance [258-260]. We consider the complexity of this DCE to be acceptable, because only 16 % of the respondents totally agreed with the statement that participation in the experiment was difficult. Reasons for experiencing this difficulty were assessing the attractiveness of an informal care situation (34%), imagining being an informal caregiver (19%), choosing between the two alternatives (61%), and reading the descriptions of the alternatives (13%). In addition, the vast majority of the study sample (85% - 92%) chose the dominating alternative of three additional 'data quality check' choice tasks[8] with dominated pairs of choices. Furthermore, respondents were familiar with the valued 'good' since almost all respondents knew what informal care was before they participated in the experiment. In addition, the number of choice sets in this study does not seem to distort the parameter estimates too much, as research showed that respondents can handle around ten choice sets [258-260].

7.4.2 CarerQol tariff

The relative utility weights of the CarerQol-7D were used to construct a tariff for the CarerQol. This tariff provides standard utility scores for caring situations described with the CarerQol-7D taking into account differences in severity of problems. In research that focuses on the burden and support of caregivers or in medical research, this tariff can be used as an outcome measure serving as a weighted sum score of subjective burden taking into account differences in dimensions of subjective burden. In addition, the tariff facilitates the inclusion of informal care in economic evaluations, explicitly on the effect side. The inclusion of the resulting utility scores in economic evaluations deserves attention, however. When the intervention is aimed at the informal caregiver (e.g., evaluating respite care), it can be used as the primary outcome measure. This would facilitate a cost-utility analysis in which the utilities refer to the informal care-related utilities. An advantage of using care-related quality of life in those contexts is that all important aspects of burden, such as fulfilment and problems combining care with other activities, are included. It also includes health effects of informal care. Hence, combining these outcome measures with QALY changes in informal caregivers brings the risk of double-counting health effects. When the CarerQol is used in the context of more conventional (patient-oriented) economic evaluations, inclusion is less straightforward. It is clear that given the conceptual differences between health-related quality of life of patients (measured in terms of QALYs) and the here central care-related quality of life (measured using the CarerQol), the two outcome measures cannot be straightforwardly combined (although both may be labelled as 'utilities'). Both measures focus on different aspects of general quality of life: that is, health for QALYs and the impact of caregiving for care-related quality of life instruments. Combining the two would require both concepts to be combined into one overall broad outcome measure, such as comprehensively measured general quality of life. This means that the CarerQol-7D cannot be included, in a straightforward manner, in the denominator of cost-effectiveness or cost-utility analysis of patient-oriented interventions. Nevertheless, it would be possible to include the CarerQol utility scores next to and separate from patient effects in a cost-consequence or multicriteria analysis. For a more elaborate discussion, see Koopmanschap et al. [25]. Paying explicit attention to informal care in economic evaluations will provide policy makers with a more comprehensive and realistic comparison of costs and effects of health care interventions and will ensure the actual adoption of the often advocated societal perspective in economic evaluations.

Our results showed that the underlying preferences for informal care situations, on which the

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[8] These additional choice sets were not included in the conditional logit regression.

CarerQol tariff is based, differed among respondents. Because of this preference heterogeneity, more advanced statistical models than the MNL model were used to accurately analyse the data. Only a part of this preference heterogeneity is explained by the interaction effect of no mental and no physical health problems in the model. Another part of the unexplained preference heterogeneity may be associated with socio-demographic factors. We tested for the interaction effects of age and gender and found, for example, that older people consider having physical health problems to be less problematic than younger persons. While this result is interesting, this chapter intentionally did not include any socio-demographic variables in the final model. The aim of this study was to built a model to construct a tariff reflecting the societal perspective, and, therefore, non-discriminative and applicable in different study settings. Again, a strategy was adopted resembling that commonly used and advocated for deriving health state preferences.

Using the CarerQol instrument and utility scores in practice, also requires further inspection of the other elements in a cost-effectiveness analysis, in order to ensure that all relevant elements are captured and double-counting is avoided. Concerning validation of the CarerQol tariff, it is important to note that although the findings in the (independent) pilot study were highly comparable to those in the main study, a topic for next research is replication of this study in another independent sample of caregivers in the Netherlands. In addition, it should be stressed that the tariff presented here reflects preferences for informal care situations of the Dutch general public and may not necessarily be transferable to other countries. Deriving tariffs for other countries remains an important area for future research.

In conclusion, the availability of a tariff for the CarerQol instrument offers the opportunity to capture the impact of informal care in economic evaluations, reflecting the preferences of the Dutch general public. Although these informal care situation-related utilities cannot be simply added to health-related utilities, this practical argument cannot justify ignoring informal care in economic evaluations. Informal caregivers are an integral part of health care systems and therefore influence the costs and effects associated with health care programs, requiring accounting for informal care within economic evaluations. The current study aims to facilitate this.

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CHAPTER

8

How to include informal care in economic evaluations

Based on Hoefman, R. J., van Exel, J., & Brouwer, W. (2013). How to Include Informal Care in Economic Evaluations. PharmacoEconomics, 31(12), 1105-1119.

Abstract

Economic evaluations of health interventions aim to support decision making in health care. To effectively do so, evaluations need to include all relevant costs and effects of an intervention. Informal care provided by family or friends is an important element of care for many patients, but can have a profound impact on the health and well-being of caregivers. Therefore, informal care should be considered in economic evaluations of health interventions. Different methods to do so exist. This chapter provides an overview of state-of-the-art methods available for this purpose, illustrated with practical examples. Since the choice of measurement and valuation technique depends on the type and perspective of the economic evaluation, this chapter supports researchers in choosing the appropriate techniques to include informal care in their economic evaluation of a health intervention. We discuss the different approaches to measuring and valuing informal care, covering both partial and full valuation methods, allowing inclusion as costs or effects.

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[9] An informal carer is defined here as a person who provides care and support to a family member, friend or acquaintance with a chronic illness, disability or other long lasting care need due to ill health or aging. Informal care is typically provided on a voluntary

8.1 Introduction

Economic evaluations of new health care technologies, especially pharmaceuticals, are increasingly used to support policy decisions in health care [70-72]. To be useful for that purpose, such evaluations need to include all relevant costs and effects in an appropriate way. While this may seem straightforward, in practice it is not. Appropriately measuring, valuing and including all different costs and effects in an economic evaluation can prove a difficult task. This is already true for aspects such as medical costs and measuring health benefits, but especially holds for costs and effects that are less central in common economic evaluations. Informal care[9] is an important example. Informal care constitutes a substantial part of the total care received by (especially chronically ill) patients and elderly in many countries [2, 3]. Informal care can complement and substitute the formal care patients receive. It can complement formal care, for example, when informal caregivers assist patients with the management of their disease or medication. This improves the overall quality of care for patients who are not fully independent (e.g., in the context of ageing, dementia, mental illness) [261, 262]. Informal care can also substitute formal care by supporting patients to stay at home longer or to be discharged from formal care sooner [66-68].

The impact of informal care on patients and caregivers can be profound [28, 42, 48-50, 159, 160]. Therefore, it is important to consider informal care in economic evaluations. This obviously holds for evaluations adopting a societal perspective. From this perspective, all relevant societal costs and effects of an intervention need to be included in an evaluation, regardless of where they fall [29, 74, 75, 77, 78]. Nonetheless, informal care is also relevant when a narrower perspective is adopted. For instance, in England and Wales where the National Institute for Health and Clinical Excellence (NICE) currently prescribes the health care perspective [114], in which only costs that fall within the health care budget and only effects on health or health-related quality of life (HR-QOL) should be considered. Then, policy makers interested in optimizing health from a given budget are likely to be interested in health changes in caregivers as well. Such health changes in caregivers have been demonstrated to exist and may have two distinct sources. First, they may be related to caregiving activities (the 'caregiving effect'). These activities can be physically and mentally straining [28, 42, 48-50, 159, 160]. Secondly, the health status of patients can directly affect their caregivers. This is labelled the 'family effect' [50, 53, 85].

Informal care is relevant in many care situations and, hence, for economic evaluations of health interventions. Although the interest in effects on caregivers appears to be increasing (which this chapter underlines), until some years ago economic evaluations often ignored informal care [92]. Common explanations for disregarding informal care include the methodological difficulties of measuring and valuing the impact of informal care [25, 91, 92]. However, numerous measurement and valuation methods of informal care have become available in recent years [104, 162], thus facilitating the inclusion of informal care in economic evaluations. Ignoring informal care is problematic, because it may result in biased calculations of cost-effectiveness and, hence, in wrong policy information and decisions. Krol and colleagues (2011) for instance, highlight the large impact that ignoring productivity costs can have on the outcomes of evaluations, which could imply the difference being considered cost-effective or not [263]. Ignoring the costs and effects of informal care may have similar impacts, especially in the context of disease areas where informal care is relatively important, such as in the case of Alzheimer's disease or rheumatoid arthritis, e.g., Brouwer et al. (1999, 2004) and Goodrich et al. (2012). Whether informal care is important in the context of any specific intervention,

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basis, arising from a prior social relationship, and without financial compensation or specific training. This definition combines several aspects from descriptions of informal care in the literature (e.g., [3, 162, 316]).

and hence needs to be included, should be an important consideration for researchers when designing their study.

This chapter aims to guide researchers in measuring and valuing the impact of informal care, in very practical terms. To this end, common methods to include informal care in economic evaluations of health interventions are summarized and practical examples of how to apply these methods are presented. To remain practical, this chapter does not provide an exhaustive, systematic review of methods and instruments, but largely draws on experience built up in the course of developing the iMTA Valuation of Informal Care Questionnaire (iVICQ) and its accompanying manual [162]. Furthermore, this chapter points out to researchers how to select the appropriate methods for their own research. It will be discussed which valuation methods are suitable per type of economic evaluation and perspective. In doing so, the focus is primarily on a commonly used type of economic evaluations: cost-utility analysis (CUA). Given the similarities of techniques, CUA and cost-effectiveness analysis (CEA) are discussed simultaneously. Some attention is also paid to the applicability of methods in cost-benefit analysis (CBA) and multi-criteria analysis (MCA). Finally, in the following sections the primary focus is on including caregiving effects in economic evaluations of patient-oriented interventions, in terms of time investment, health or well-being effects. However, some of the methods discussed can also be used when evaluating support interventions aimed directly at informal caregivers. Moreover, they may be used to measure and value the family effects caregivers (and other non-caring family members) may experience.

8.2 How can informal care be measured?

Informal care can be included in the numerator or denominator of an incremental cost-effectiveness ratio (ICER). In other words, informal care can be captured on the cost-side or on the effect-side of an economic evaluation.

8.2.1 Measurement of the costs of informal care

Two main types of costs of informal care can be distinguished: (i) out-of-pocket expenses, such as travel expenses, and (ii) time input of caregivers, also called the objective burden of caregiving. Out-of-pocket expenses can be measured by directly asking caregivers about expenses for informal care. Time input by caregivers typically is more important. Researchers can use different methods to measure time input, such as the diary method and the recall method [16, 265]. The diary method is often considered to be the preferred method for recording time use of individuals. However, registering all activities performed in a specific timeframe is very time consuming. Moreover, this method can be straining for respondents and, hence, may not always be feasible [16]. With the recall method, researchers retrospectively ask about the number of hours spent on care tasks during the last week, such as for example in chapter 3. An example of such a question is shown in Figure 8.1. This method provides a valid measure of time input as compared to the diary method [16].

If desired, more detailed questions focusing on specific activities are available for examples, see previous publications [11, 13, 50, 53, 110, 266, 267]. These questions provide more insight in the specific activities performed by caregivers. Furthermore, the number of tasks performed shows the complexity of caregiving situations, which can be an important determinant of the experienced subjective burden of caregiving [34].

How much time during the last week did you spend on household activities that would not have had to be performed if she/he were in good health, or if she/he could have done them?

For example, food preparation, cleaning, washing, ironing, sewing, taking care of and playing with your children, shopping or maintenance work, odd jobs, gardening.

______________________________ hours during the last week

How much time during the last week did you spend on personal care for her/him?

For example, dressing/undressing, washing, combing, shaving, going to the toilet, mobility around the house, eating and drinking and medication.

______________________________ hours during the last week

How much time during the last week did you spend on practical support that would not have had to be performed if she/he were in good health, or if she/he could have done it?

For example, mobility outside the house including assistance with walking or wheelchair, visiting family or friends, seeing to health care contacts (e.g , doctors' appointments), organising help, physical aids or house adaptations and taking care of financial matters (e.g., insurance).

______________________________ hours during the last week

Figure 8.1 Example of recall method for measuring time input by informal caregivers [162]

Accurate time registration in the context of informal care often is problematic. Measurement bias may arise for instance from joint production, e.g., doing two things at the same time, such as surveillance while doing normal household work. Bias may also arise from difficulties in separating time spent on normal tasks and informal care tasks, e.g., caregivers that took over household tasks from the patient years ago may now see this as normal time use rather than informal care [16]. Moreover, sometimes respondents indicate that they spend 24 hours per day on caring, for seven days a week. This obviously is not realistic, because caregivers also need time for personal care and sleep. Such an answer therefore seems an expression of strain and involvement rather than an accurate registration of sacrificed time use. In such cases, previous studies have, for example, set a limit of 18 hours per day (or 126 hours per week) for the total number of hours caregiving for examples, see previous publications [28, 100] or chapter 3. Caregivers are then still assumed to spend most of their time on care activities, like they indicate themselves, but also to have an average of 6 hours per day for their own basic needs. The validity of registered time inputs should always be considered carefully. Since a golden standard is lacking, this could be done by within-study checks (e.g., by linking number of hours to patient status) and by across-study comparison, preferably with studies using a similar patient/caregiver population and a comparable institutional context. It would also be helpful if more review studies became available in this area, like those reported for dementia [268, 269].

8.2.2 Measurement of the effects of informal care

The impact of informal care can be quantified as an effect by measuring caregivers' (i) subjective burden (ii) health or (iii) well-being. Subjective burden is the strain of caregiving as perceived by caregivers. This subjective burden is not necessarily strongly related to the objective burden of caregiving [25, 29, 34]. Put differently, some caregivers perform many care tasks but do not feel strained, while others perform just a few tasks and consider this to be very straining. A variety of subjective burden instruments exist. Most provide a detailed description of the impact of caregiving on things like mental health, physical health, and social and financial problems [26, 104, 105]. Some instruments implicitly or explicitly include positive aspects of caring, such as fulfilment from caring, e.g., the Self-Rated Burden scale (SRB; [29, 105]) and Caregiver Strain Index plus (CSI+; [33, 107]). It is important to note that subjective burden instruments do not provide an economic valuation of informal care. Therefore, these instruments are unsuited to include informal care in the most common types of economic evaluations. This is also the case for some instruments that have been developed to measure caregiver quality of life (see, for example Deeken et al. (2003)), but lack an aggregate utility score. How the information such instruments provide can be used in the context of economic evaluations is highlighted in section 4.

The effect of providing informal care on caregivers' health can be expressed in changes in health-related quality of life (Table 8.1) [13, 29, 50, 53, 54] and measured in terms of Quality-Adjusted Life Years (QALYs; Table 8.1, e.g., [74]). In the context of an economic evaluation, this can best be assessed directly by measuring changes in caregivers' health due to the intervention. In other words, measuring differences in health of caregivers between treatment arms. If this is not feasible, one can fall back on indirect evidence. For instance, by investigating the influence of informal care hours on health [50]. Alternatively, researchers can compare health of caregivers to the health of the population at large [13, 270]. It needs noting, however, that health effects may originate not only from the strain of caregiving, but also from the mere fact that a loved one has a serious illness or condition. This family effect can occur in a broader range of significant others than just the carer(s) [50, 53].

Informal care can have an impact on different life domains, also beyond health. Therefore, one may also consider measuring well-being, or general quality of life, of caregivers. One way of doing so is by measuring happiness of caregivers. Happiness, in general terms, is the judgment of an individual of the quality of their life as a whole [125]. As such, it can be seen as an expression of (experienced) utility or welfare, relevant in economic decision making. Important to note here is that changes in happiness may have different underlying sources. For example, caregiving can influence happiness of caregivers, but the knowledge that a direct family member suffers from a serious illness or condition can also have an impact, i.e., the family effect. Therefore, as with health effects, relevant changes in well-being can occur in a wider range of significant others, e.g., non-caring family members [53].

Table 8.1 Overview of non-monetary valuation methods of informal care

Concept	Valuation method
Health-related quality of life	Quality-Adjusted Life Year caregiver (QALY cg)
Care-related quality of life	Carer Experience Scale (CES) Care-related Quality of Life -7 Dimensions (CarerQol-7D)
Well-being	Care-related Quality of Life - Visual Analogue Scale (CarerQol-VAS) Process Utility (PU)

8.3 How can informal care be valued?

Inclusion of informal care in economic evaluations requires a valuation in economic terms of the measured inputs or effects. Different options are available for the valuation of informal care [25, 91, 92]. The principal differences relate not only to what is valued, but also to how the valuation is performed. A first important choice is whether one wishes to incorporate informal care in the numerator or the denominator of the ICER. In other words, researchers need to choose whether they will capture informal care on the cost or effect side of an economic evaluation. Monetary valuation methods value informal care costs, non-monetary valuation methods value carer effects. Moreover, methods differ in the range of consequences of informal care they consider in the valuation. Valuation methods can provide either a partial or a full valuation of informal care. A partial valuation focuses only on a selection of consequences of caregiving, such as time investment. Full valuation methods of informal care include all consequences of caring. In general, from a welfare economic viewpoint, full valuations of informal care are preferred in economic evaluations. However, when an evaluation uses a narrower perspective, partial valuation methods may be required.

8.3.1 Monetary valuation of informal care

Monetary valuation methods express the value of informal care in monetary terms. This value is multiplied with the number of care hours and included on the cost side of an economic evaluation. Normally, one derives a monetary value of a product or service from its market value. In other words, by using the observed prices. However, informal care is not traded on a normal market. Therefore, one cannot directly observe market prices for informal care [25, 26, 29, 96, 264, 271]. Hence, estimating the value of an hour of informal care requires other methods. Several of these methods exist: the opportunity cost (OC), proxy good (PG), well-being (WB), contingent valuation (CV), and conjoint analysis (CA) methods (Table 8.2). These methods differ in the way to derive values. This is explained in section 8.5.

The OC and PG methods provide a partial valuation of informal care. Both methods only consider the value of the time caregivers sacrifice. Moreover, the OC and PG method value all hours equally. However, the value may differ between hours. That is, people may value the first hour different than the 20th hour of care. Moreover, people may prefer performing specific care tasks over others. Such preferences are not reflected in these methods [25, 26, 91]. The WB, CV and CA methods in principle[10] provide a full valuation of the impact of informal care.

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[10] These methods can also partially value informal care, depending on the valuation exercise, e.g., when a willingness-to-pay question specifically excludes health or labor participation effects, the valuation is clearly partial when such effects do occur.

An important advantage of valuing informal care in monetary terms is the straightforward and uncomplicated inclusion in economic evaluations. The derived costs (or savings) can simply be added to other costs. A downside is that adding of the costs of informal care to the stack of other cost items in economic evaluations gives less explicit attention for the consequences of a health care program on caregivers [110]. Non-monetary valuation of informal care, which is discussed in the next section, may provide more explicit insight in the exact consequences of informal care.

Table 8.2 Overview of monetary valuation methods of informal care

Concept		Valuation method
Revealed preference based methods		
	Time input caregiver	Opportunity cost method (OC) Proxy good method (PG)
	Well-being caregiver	Well-being method (WB)
Stated preference methods		
	Willingness to pay (WTP) / Willingness to accept (WTA)	Contingent valuation (CV) Conjoint analysis (CA)

8.3.2 Non-monetary valuation of informal care

The consequences of informal care, which may be negative as well as positive, can be made more explicit in an economic evaluation by expressing it as an effect [264]. Moreover, doing so may be perceived to be consistent with the common division between costs and effects, at least for certain aspects of informal care (especially health effects). Existing non-monetary valuation methods (Table 8.1) value effects in terms of caregivers' health-related quality of life (using QALYs), care-related quality of life, or well-being (happiness). By focusing on a single dimension of quality of life, i.e., the health domain, QALYs comprise a partial valuation of informal care [25, 26]. Measures for deriving QALY changes are readily available from patient studies (e.g., EQ-5D, SF-36, etc.). Care-related quality of life conceptually resembles health-related quality of life valuations commonly used in economic evaluations (in terms of QALYs), but values a broader range of utility impacts than only health. The focus is typically on the most prominent impacts of informal care on general quality of life, both negative and positive. Care-related utility scores can be calculated for each possible care profile using tariffs. Hence, researchers can derive changes in care-related utility of caregivers. Two care-related quality of life measures are currently available: the Carer Experience Scale (CES) [113] and the Care-related Quality of Life instrument (CarerQol; chapters 2-7, [110]), see section 8.6.3. These measures aim to provide a full valuation of informal care. They focus on capturing the effect of informal caregiving on well-being. Therefore, factors not directly related to caregiving, such as wealth, are less likely to influence such care-related quality-of-life scores. The general well-being score is also increasingly used as an outcome measure in the field of (health) economics [123, 124]. The main advantage of this broad valuation measure is that all the different effects of informal care, such as health effects, financial problems or fulfilment from caregiving, are taken into account through their impact on general well-being. Therefore, well-being scores can constitute a full valuation of informal care.

However, effects outside the direct scope of caregiving, such as educational level, type of job, and family life, might influence well-being answers as well. Moreover, the same applies to coping of caregivers to their stressful situation [26]. Hence, the way in which one derives well-being estimates is important. This is emphasized by the fact that the causality of the relationship between caregiving and well-being effects is ambiguous [25].

Sections 8.5 and 8.6 provide more detailed information on the application of both the monetary and non-monetary valuation methods. First, however, it is highlighted how informal care can be included in different types of economic evaluations.

8.4 How can informal care be included in cost-effectiveness/-utility analysis?

8.4.1 Cost-effectiveness and cost-utility analysis

Whether and how informal care can be included in economic evaluations importantly depends on the type of economic evaluation that is conducted. In this section, it will be discussed which valuation methods can be used to include informal care in CEA or CUA. Moreover, preferred options for including informal care in these types of economic evaluation are proposed.

CEA and CUA can be conducted from a health care or societal perspective. The perspective determines which costs and effects are relevant to include in the cost-effectiveness calculations. Consequently, this affects which valuation methods are appropriate. A study conducted from a health care perspective can include health effects (QALYs) in caregivers on the effect side (Table 8.3). This health information of caregivers can easily be aggregated with patient QALYs at the effect side of the CEA/CUA. Important to note here is that this only applies if the measurement and valuation methods of health effects in patients and caregivers are similar. Hence, in such cases, researchers are advised to use the same generic health measure for the carer as used for the patient to increase comparability and possibility of aggregating effects in patients and caregivers. More research into the sensitivity of these instruments in caregivers remains important.
Costs falling on caregivers are commonly deemed irrelevant when adopting a health care perspective. Such costs occur, for example, due to sacrificed labour time of caregivers. Hence, besides health changes, typically no further valuation methods need to be applied when taking a health care perspective. However, in a CEA/CUA conducted from a societal perspective all (sufficiently large) consequences of caregiving are relevant (Table 8.3). Hence, researchers can combine health effects measured in QALYs at the effect side with time input of caregivers measured with the OC or PG method on the cost side. Alternatively, the full impact of informal care can be included at the cost side, e.g., using the willingness-to-pay (WTP)/willingness-to-accept (WTA) method (Table 8.3). It is not possible to combine QALYs with the WTP/WTA method. In principle, the latter method provides a full valuation of informal care. Therefore, WTP/WTA should already value the health effects of caregivers. Hence, combining both methods could result in double counting of health effects.

Important to note here is that researchers in general need to be aware of the risk of double counting the impact of informal care in economic evaluations when using different valuation methods. That is, if a particular consequence of informal care is already included in the numerator, it should not also be included in the denominator. Likewise, important impacts should not go unnoticed. An extensive discussion of this topic can be found in the literature [25, 26, 29, 91].

It should be emphasized that the above pertains to economic evaluations of interventions aimed at patients, in which case informal caregivers invest time in care and experience—both negative and positive—effects from caregiving. On top of these effects, as discussed in section 1, informal caregivers— and other non-caring family members—may experience family effects from the fact that their loved one is ill. The latter effects are rarely considered in economic evaluations. On the other hand, when evaluating interventions specifically aimed at informal caregivers, caregiver outcomes are central. This, for example, applies to comparative research of different types of support or respite programs for caregivers. In such cases, the overall impact of caring can be included at the effect side of a CEA/CUA using a care-related quality-of-life instrument. This provides a full valuation of the impact of informal care in effect terms.

Table 8.3 Preferred valuation method for including informal care in economic evaluations per type of economic evaluation

Economic evaluation	Perspective	Preferred valuation method
CEA/CUA		
	Health care	QALY cg
	Societal	PG/OC method + QALY cg
CBA		
	Health care	QALY cg [a]
	Societal	WTA/WTP
MCA		
	Health care	QALY cg
	Societal	Care-related quality of life

[a] Expressed in monetary terms; use same value for health effects in caregivers as used in economic evaluation for patients' health effects
CEA Cost-Effectiveness Analysis, *CUA* Cost-Utility Analysis, *CBA* Cost-Benefit Analysis, *MCA* Multi-Criteria Analysis, *QALY* Quality-Adjusted Life Year, *cg* caregiver, *PG* Proxy Good, *OC* Opportunity Costs, *WTA* Willingness to Accept, *WTP* Willingness to Pay

8.4.2 Other types of economic evaluations: CBA or MCA

A CBA performed from a health care perspective can include informal care by measuring health effects in caregivers. Because CBA expresses all costs and effects exclusively in monetary value, these health effects should be valued in monetary terms. Arguably, the same monetary value as used for monetizing health effects in patients should be used (Table 8.3). Researchers conducting a CBA from a societal perspective could opt to measure time input of caregivers and multiply this with a WTA/WTP estimate per hour of informal care (Table 8.3). If the underlying valuation exercise was sufficiently broad, this method values the full impact of informal care. MCA from a health care perspective can include informal care in its analysis by measuring QALYs of caregivers. In MCA from a societal perspective, researchers can pay explicit attention to the overall impact of a patient intervention on caregivers. This can be done by using care-related quality-of-life instruments, such as the CES or the CarerQol. The outcomes can then be presented next to other outcomes of interest. Important to note here is that researchers can also present information on the objective burden (i.e., number of hours of caregiving) or subjective burden (e.g., felt strain of caregiving) of caregiving in MCA. Different methods to measure these two types of burden are described in the iVICQ [162].

8.5 How can informal care be valued in monetary terms?

In this section, which follows on section 8.3.1, detailed guidance on how to use different monetary valuation methods will be provided.

8.5.1 Opportunity cost method

The number of hours of activities sacrificed in order to be able to provide care are central in the OC method. The OC method calculates the value of informal care by multiplying these hours of sacrificed activities with a value per hour for each activity. A general typology of sacrificed activities includes paid work, unpaid work (i.e., voluntary work or housekeeping), and leisure time (Table 8.4). To register the amount of time sacrificed per activity, recall methods can be used (Figure 8.2). A difficulty of such retrospective questions is that respondents might find it hard to indicate how much time they have sacrificed. Moreover, they might find it difficult to distinguish between 'normal time use' and informal care activities. This especially applies to caregivers who have been performing care for longer periods of time. In this situation, researchers can use hypothetical questions (Figure 8.3). In these questions, respondents indicate which activities they would perform if informal care was not needed anymore.

Table 8.4 Information needed for calculating monetary valuation of informal care

Method	Measurement of time spent on:	Valuation hour informal care based on:
OC		
	Paid work Unpaid work Leisure time	Gross personal income caregiver
PG		
	Household activities Personal care Practical support	Tariff market substitute household activities [a]
WB		
	Informal care	Conduct WB study among own sample Monetary value WB study [a]
WTP/WTA		
	Informal care	Elicit WTP/WTA in own sample with CV/CA experiment WTP/WTA from CV/CA study [a]

[a] Use external references

OC Opportunity Costs, *PG* Proxy Good, *WB* Well-Being, *WTA* Willingness to Accept, *WTP* Willingness to Pay, *CV* Contingent Valuation, *CA* Conjoint Analysis

Once the number of hours sacrificed per activity is known, these hours need to be valued. The value per hour for paid work, unpaid work, or leisure time can be set at different levels (Table 8.4). Often, the gross hourly wage of the respondent is used for paid work. However, many caregivers do not have a paid job, e.g., because they are the housekeeper or retired, and their wage rate is therefore unknown. For caregivers below retirement age, researchers can resort to an equivalent of the wage rate, such as the average hourly wage rate of persons of the same sex, age, and educational level. Another option is the reservation wage rate, which represents the hypothetical wage rate for which the unemployed carer would be willing to provide an hour of paid work. For caregivers who are retired, the activities sacrificed may be voluntary work or leisure time. If available, local tariffs from value of time studies can be applied. Otherwise, the PG method (see next section) may be the next best alternative. The use of wage rates in the valuation of informal care obviously may raise equity questions, because the (social) value of informal care is then related to the occupational position or educational level of the caregiver. Whether it is appropriate and fair that the value of an hour of informal care provided by a lawyer would be worth more than that of a waiter, is open for debate. Sometimes studies use the wage rate for all time components. Researchers can use an adapted gross hourly wage or a direct valuation to value household activities and leisure. However, both may prove difficult to obtain. Moreover, adjustments to the gross hourly wage rate tend to be relatively arbitrary. For an example of the application of the OC method, see Figure 8.4. More background information on the OC method can be found in [25, 26, 91, 99]. Monetary values of studies using the OC method to value informal care range from €5 to €29 per hour[11] (for examples, see previous publications [99, 272-286]).

Did you completely or partly give up **paid work** in order to provide informal care to her/him?

☐ No, I did not have paid work before

☐ No, I still perform the same amount of paid work

☐ Yes, for ____________________ (number) fewer hours per week

since ______________________ (year)

Did you completely or partly give up **unpaid work** in order to provide informal care to her/him?

☐ No, I did not have unpaid work before

☐ No, I still perform the same amount of unpaid work

☐ Yes, for ____________________ (number) fewer hours per week

since ______________________ (year)

Did you give up **time spend on leisure** in order to provide informal care to her/him?

☐ No, I did not have time for leisure before

☐ No, I still spend the same amount of time on leisure

☐ Yes, for ____________________ (number) fewer hours per week

since ______________________ (year)

Figure 8.2 Recall method: opportunity cost method [162]

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[11] Values are in 2012 euros. Values in euros before 2012 can be converted to 2012 euros by multiplying with annual average rates of change in harmonized indices of consumer prices (HICPs) ([317]. Currencies other than euros were first converted with historical currency rates.

Suppose you did not have to provide informal care anymore. How would you spend this time: on paid work, unpaid work, or leisure?

More paid work: ☐ No

☐ Yes: ____________________hours per week

More unpaid work: ☐ No

☐ Yes: ____________________hours per week

More leisure: ☐ No

☐ Yes: ____________________hours per week

Figure 8.3 Hypothetical questions: opportunity cost method [162]

Suppose a respondent provides 12 hours of informal care per week, giving up the following to provide informal care:

- 1 hour paid work
- 3 hours unpaid work
- 8 hours leisure time

If the respondent's gross hourly wage rate is €30, the value of unpaid work and leisure time is set to the value of household activities of €12.50 per hour, then the monetary value of the time forgone to provide informal care of this person is (1 * 30) + (3 * 12.50) + (8 * 12.50) = €167.50.

Figure 8.4 Numerical example: opportunity cost method [162]

8.5.2 Proxy good method

The PG method calculates the value of informal care by multiplying the number of hours spent on informal care by a value per hour for each care task performed. Usually household activities, personal care and practical support are distinguished (Table 8.4). In the PG method, the value per hour is based on the shadow price of a market substitute. Market prices can vary per task, because they are performed by care professionals earning different wages (Table 8.4). Several studies have applied the PG method. Values per hour spent on informal care in these studies range from €3 to €26^{11} [99, 272, 280, 287-293]. For example, in The Netherlands the tariff for a market substitute for household activities is set to €12.50 [294].

The PG method uses wages earned by substitutes such as household helpers or specialized nurses. In doing so, the PG method provides insight into the costs of replacing informal care with formal care. An important, and probably unrealistic, assumption underlying this method is that formal and informal caregivers are perfect substitutes. This would imply that they can take over each other's tasks without efficiency or quality losses (or gains). For instance, by using the wage rate of a professional nurse, one implicitly assumes that a nurse spends the same amount of time on an activity as a caregiver. Contrary to the OC method, the PG method does not relate the value of informal care to the occupational position or educational level of the carer. The PG method, and its pros and cons, are extensively discussed in previous publications [25, 26, 91, 99] and the method is applied by Van den Berg et al. (2006). Figure 8.5 presents an example of the PG method.

Suppose a respondent provides 12 hours informal care per week, comprising:

- 7 hours household activities
- 2 hours personal care
- 3 hours practical support

If the shadow price of household activities is €8.50, personal care €35, and practical support €35, then the monetary value of the time forgone to provide informal care is (7 * 8.50) + (2 * 35) + (3 * 35) = €234.50.

Figure 8.5 Numerical example: proxy good method [162]

8.5.3 Well-being method

Central in the WB method is the change in well-being of the carer due to caregiving. The WB method calculates the monetary amount required to compensate a carer for her/his loss in well-being due to caregiving. The WB method multiplies this monetary value of an hour of caregiving with the number of hours spent on informal care (Table 8.4). For instance, suppose that the happiness of a carer drops from 8 to 7 on a scale from 0 to 10 due to straining caregiving tasks. Assuming that income positively influences well-being, the WB method can derive the increase in income that would exactly offset the drop in well-being caused by caregiving. In other words, the income increase leading to a 1-point increase in well-being. While obtaining well-being scores in population samples is possible [119] and is becoming more common, transforming these into a monetary valuation of informal care requires substantial research effort. In most cases, it will not be possible to perform this method within an economic evaluation. Alternatively, it is possible to use previously obtained values. Researchers then multiply these values with the number of hours of informal care as measured within the economic evaluation. Depending on the availability in the literature, values could be differentiated according to the intensity of caregiving or tasks performed. Notes on the calculation of a monetary value of informal care using this method can be found in Van den Berg et al. [91]. Van den Berg and Ferrer-i-Carbonell (2007) applied the WB method and reported an average monetary value of approximately €12^{11} per hour of informal care.

8.5.4 Stated preference methods

8.5.4.1 Contingent valuation and conjoint analysis

The CV and CA methods calculate the value of informal care by multiplying the number of hours spent on informal care by a value per hour (Table 8.4). CV derives this value by presenting respondents with a hypothetical caregiving situation and asking them to specify the minimum compensation they would require for providing an extra hour of informal care (Figure 8.6)[12]. This is an example of the WTA approach. Using CV, one can also ask what the maximum amount is that respondents are willing to pay to reduce their informal care provision with 1 hour (WTP). In the context of valuing informal care WTA seems the more appropriate approach, because the common (policy) perspective of valuation questions is one of increasing caregiving and thus giving up time on other activities. Following this perspective, asking respondents about the minimum compensation required to provide an additional hour of care seems conceptually most appropriate [295, 296]. Nonetheless, WTP is more commonly used in valuation studies, often with reference to recommendations of the authoritative NOAA (National Oceanic and Atmospheric Administration) Panel that advocated WTP as the preferred approach [297].

In CA, or a discrete choice experiment (DCE), the monetary value of an hour of informal care is derived from respondents in an indirect manner [245]. In these experiments, respondents choose between two or more hypothetical informal care situations. These situations are described by different characteristics, also called attributes. These are, for example, the number of hours of care per week and the type of care tasks. To derive a monetary value, one of the attributes should concern money. For instance, an hourly wage rate received for informal care or a certain tax refund from government can be used. By varying the levels of the attributes and having respondents make several choices, implicit preferences for attributes can be derived. Using this information, researchers can calculate the monetary value of all attributes.

Imagine that she/he needs **one extra hour of informal care** per week and that government will pay you for lending this extra hour of informal care. Please look at the numbers below, from left to right, and tick the *highest* amount that you **would definitely not** be willing to forgo to provide an extra hour of informal care.

For example: if you are certain that you would not provide the extra hour of informal care for €20 from the government, but not certain that you would forgo €22.50, tick €20.

0	5	7.50	10	12.50	15	17.50	20	22.50	25	30	35	40	45	50	higher
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

Figure 8.6 Example of willingness to accept question for informal care (incomplete version) [162]

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[12] In these experiments, only the number of hours per week change; all other things, such as the recipient's need for care, are assumed to remain equal in order to avoid contamination of values with other aspects (such as the health of the patient).

Responses to stated preference methods such as CV and CA are known to be prone to different biases, including strategic answering, starting point-bias, and hypothetical bias. Moreover, in the case of CA, it can be quite difficult for respondents to compare different (hypothetical) scenarios. Finally, especially when using WTP/WTA techniques, one needs to be aware of the fact that some respondents may find it unethical to receive money to provide more informal care or pay money to reduce it [26, 267]. For a discussion of the various biases in CV, see Van Exel et al. (2006).

Researchers can obtain monetary values in the study sample within the context of an economic evaluation. It is often more feasible for researchers to conduct CV than CA in an economic evaluation. That is, designing a CA study tends to be more complex and respondent burden is often higher. Below an (incomplete) example of a WTA question is provided, which can be used in this context. The complete WTA exercise is included in the iVICQ [162]. More information on informal care CV studies can be found in the literature [117, 267, 296, 298]. Practical applications of DCEs to the monetary value of informal care can be found elsewhere [113, 118, 255-257]. When obtaining values is not feasible in a specific study, researchers can use values reported in the literature. Monetary values for an hour of informal care found in these studies range from €4 to €14[11] for CV experiments [267, 299, 300] and from €1 to €15[11] in CA studies [118, 255, 257].

8.6 How can informal care be valued in non-monetary terms?

In this section, which follows on from section 8.3.2, a more detailed guidance on how to use different non-monetary valuation methods will be provided.

8.6.1 Health of caregiver

The health status of caregivers in terms of QALYs can be measured with validated health utility instruments such as the EQ-5D [122] or SF-6D [301] and valued using national tariffs [130, 131], as is usually done in economic evaluations (Table 8.5). Preferably, researchers use the same generic health measure for the caregivers as the patient. This will increase comparability and possibility of aggregating effects in patients and caregivers. Ideally, researchers observe health effects in the context of a randomized controlled trial (RCT).

We discourage using predefined algorithms assuming some stable relationship between the health of caregivers and patients. The relationship between carer health and patient health is unlikely to be stable [26]. Often, if the patient's health improves, the health of the carer will also improve through reduced informal care needs. However, this need not be the case. For example, when the health of the patient improves this could postpone institutionalization of the patient leading to prolonged informal care provision. Moreover, the relationship between patient and carer health does not need to be linear or symmetrical [26].

Table 8.5 Information needed for calculating non-monetary valuation of informal care

Concept	**Instrument to measure and value**	
Health-related quality of life caregiver	Health utility instrument	Tariff to calculate QALYs [a]
Care-related quality of life	CES	Tariff to calculate care-related quality of life [a]
	CarerQol-7D	
Well-being caregiver	CarerQol-VAS	
	Process Utility	

[a] Use external references; CES tariff can be found in Al-Janabi et al. [117], tariff for the CarerQol-7D can be found in chapter 7 and in Hoefman et al. [168]
CES Carer Experience Scale, *CarerQol* Care-related Quality of Life, *QALY* Quality-Adjusted Life Year

8.6.2 Well-being of caregiver

The CarerQol-Visual Analogue Scale (VAS) (Table 8.1), values informal care in terms of well-being changes in caregivers. The CarerQol-VAS is part of the CarerQol instrument [110]. The CarerQol-VAS is a horizontal VAS ranging from completely unhappy (with the value 0) to completely happy (with the value 10). Researchers can observe differences in well-being in the context of the evaluation of some intervention by comparing the caregivers in the two (or more) arms of an RCT.

Researchers can use an additional question to the CarerQol-VAS to quantify the process utility (PU) from caring (Table 8.1). PU refers to the value attached to the process of lending informal care by caregivers [28]. PU provides insight in the desirability of other persons taking over (all) care tasks. To calculate PU, the difference in happiness between two situations is taken: the CarerQol-VAS score of the current situation minus the CarerQol-VAS score of a hypothetical situation. In this hypothetical situation, a person selected by the care recipient and caregiver would take over the care tasks, without changing the living situation of the care recipient and free of charge.

8.6.3 Care-related Quality of Life

To date, two measures of care-related quality of life of caregivers that allow utility measurement are available. Both are highlighted in the following sections.

8.6.3.1 Carer Experience Scale

The CES contains six dimensions of caregiving: (i) activities outside caring, (ii) support from family and friends, (iii) assistance from organizations and the government, (iv) fulfilment from caring, (v) control over the caring, and (vi) getting on with the care recipient. Respondents score their care situation, by indicating the level of problems on these six dimensions. Based on the profile indicated, a care-related utility value can be attached to the profile, using a tariff based on preferences of caregivers of elderly persons in the UK. This tariff ranges from 0 (worst caring state) to 100 (best caring state) (Table 8.4). Instructions for calculating the CES score and more general information on the use of the CES can be found in Al-Janabi et al. (2008, 2011).

8.6.3.2 CarerQol instrument

The CarerQol instrument includes the CarerQol-VAS (discussed in section 8.6.2) and the CarerQol-7 Dimensions (7D). The CarerQol-7D consists of five negative and two positive dimensions of informal care. Negative dimensions are (i) relational problems, (ii) mental health problems, (iii) problems combining daily activities with care, (iv) financial problems, and (v) physical health problems. The positive dimensions are (vi) fulfilment from caregiving and (vii) support with lending care. As shown in Figure 8.7, respondents indicate whether an item applies to them with three possible responses: (i) no, (ii) some, and (iii) a lot. Answers on the negative dimensions of the CarerQol-7D receive value of 0 (a lot), 1 (some), and 2 (no). Answers on the positive dimensions receive a value of 0 (no), 1 (some), and 2 (a lot). The CarerQol has been applied in several studies (chapters 2 and 3, [54, 100, 110]). A care-related quality-of-life score can be derived from the CarerQol-7D profiles, using a tariff based on preferences from the general population in The Netherlands [162] (Table 8.5). These tariffs are reported in the iVICQ [162] and yield care-related utility scores ranging from 0 (worst informal care situation) to 100 (best informal care situation) (Figure 8.8). In the iVICQ, syntax files for SPSS® and Stata® are provided to calculate CarerQol-7D scores.

We would like to form an impression of your caregiving situation.
Please tick a box to indicate which description best fits your caregiving situation at the moment.

Please tick only one box per description: 'no', 'some' or 'a lot of'.

	no	some	a lot of	
I have	☐	☐	☐	fulfilment from carrying out my care tasks.
I have	☐	☐	☐	relational problems with the care receiver (*e.g., he/she is very demanding or he/she behaves differently; we have communication problems*).
I have	☐	☐	☐	problems with my own mental health (*e.g., stress, fear, gloominess, depression, concern about the future*).
I have	☐	☐	☐	problems combining my care tasks with my daily activities (*e.g., household activities, work, study, family and leisure activities*).
I have	☐	☐	☐	financial problems because of my care tasks.
I have	☐	☐	☐	support with carrying out my care tasks, when I need it (*e.g., from family, friends, neighbours, acquaintances*).
I have	☐	☐	☐	problems with my own physical health (*e.g., more often sick, tiredness, physical stress*).

How happy do you feel at the moment?

Please place a mark on the scale below that indicates how happy you feel at the moment.

Figure 8.7 Care-related Quality of Life instrument (CarerQol) [110, 162]

Suppose that the answers of a respondent on the CarerQol-7D are:

- *some* fulfilment
- *a lot of* relational problems
- *no* mental health problems
- *some* problems combining daily activities
- *no* financial problems
- *a lot of* support
- *no* physical health problems

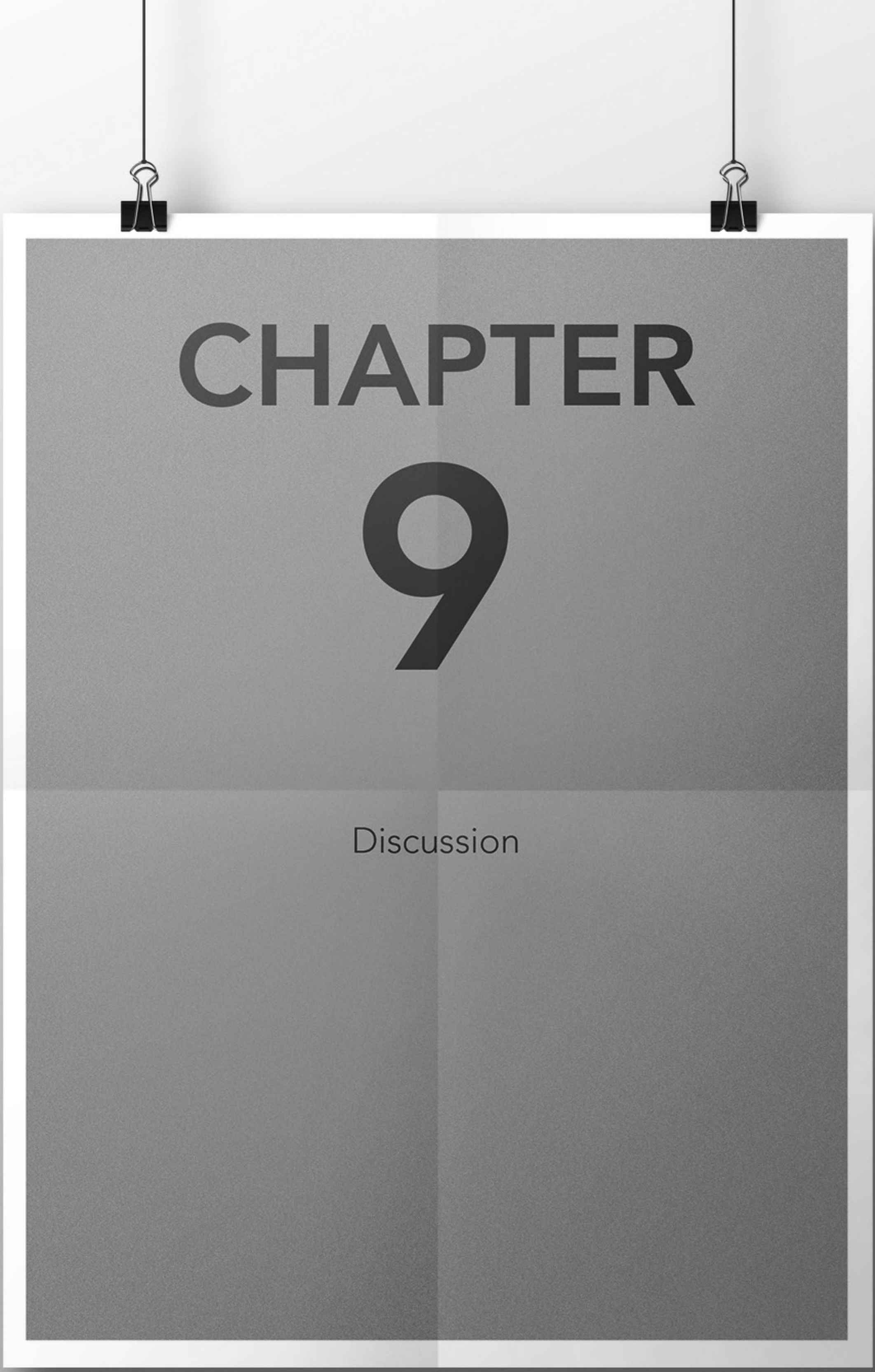
The CarerQol-7D score is: 13.6 + 0 + 13.3 + 6.4 + 14.3 + 6.5 + 15.1 + 6.6 = 75.8

It is important to keep in mind when calculating the CarerQol-7D score that respondents get a 'bonus' of 6.6 for having neither mental nor physical health problems.

Figure 8.8 Numerical example of the Care-related Quality of Life - 7 Dimensions (CarerQol-7D) score [162]

8.7 Conclusion

Including informal care in economic evaluations of health care interventions poses important methodological questions. However, several options are available for researchers to include it in a suitable way. This chapter has highlighted these options and highlighted methods that facilitate inclusion in a fairly straightforward way. We strongly encourage researchers to include informal care in economic evaluations. This will allow decision makers to be fully informed about the costs and consequences of health care interventions, not only in patients, but also in their caregivers. Given the impact informal care can have on the lives of caregivers and its important role in the health care sector, providing information on this impact to policy makers is clearly important. Instruments such as the iVICQ [162] provide further guidance on how to do so. More research in this important field and increased consensus on how to value informal care in practice remains important.



CHAPTER

9

Discussion

9.1 Introduction

Family and friends fulfil an essential role in addressing the care needs of people confronted with illness, disability or infirmity of old age. Any changes in formal health care services provided within a country may not only affect patients, but also their informal caregivers. Unfortunately, policy makers are usually ill informed about the consequences for caregivers of such changes, including those following from decisions to reimburse or –in particular- not reimburse particular treatments. An important reason for this is that economic evaluations comparing the costs and effects of health care interventions typically do not include the costs and effects of informal care. This neglect of the impact of caregiving on caregivers may consequently result in biased cost-effectiveness recommendations and lead to decisions that will not improve the welfare of society as a whole and that, in particular, may negatively impact the welfare of informal caregivers.

For a long time, methodological challenges in quantifying the costs and effects of informal care have complicated the inclusion of informal care in economic evaluations. For example, measurement tools documenting the strain from caregiving typically focus on negative aspects of informal care only, passing by the positive aspects of caregiving which motivate caregivers to start and continue their care task, and that may at least in part alleviate their strain. Consequently, these instruments only record a part of the wide spectrum of consequences of providing care on caregivers. In addition, most of these instruments are restricted to measuring problems with caregiving without attending to differences in how these problems are experienced by individual caregivers, and therefore may fail to inform policy makers fully of the actual consequences of caregiving on caregivers.

The central goal of this thesis was to overcome some of these methodological difficulties, in order to better enable inclusion of informal care in economic evaluations. In this context, three issues were studied. The first concerned investigating the psychometric properties of an instrument designed to measure the full impact of providing care on caregivers for inclusion in economic evaluations, the CarerQol instrument [110]. Secondly, this thesis presented tariffs based on preferences of the general public for the caregiving states described with the CarerQol instrument. These preference-based tariffs therefore take into account differences in the importance of caring problems. Finally, this thesis provided an overview of currently available methods to measure and value informal care for use in economic evaluations. Accordingly, the following three research questions were addressed in this thesis:

1. What is the feasibility, test-retest reliability, and construct validity of the CarerQol instrument as a measure of the effect of caregiving on caregivers in different caregiving contexts;
2. What are the preferences of the general public in the Netherlands for caregiving situations described by the CarerQol instrument;
3. How can informal care be included in economic evaluations of interventions in health care?

This final chapter discusses the main findings of this thesis and the limitations in answering each of the research questions. The chapter ends with a general conclusion, suggestions for future research and policy recommendations.

9.2 Psychometric properties of the CarerQol instrument

Five studies included in this thesis (chapters 2 - 6) addressed the feasibility, test-retest reliability and construct validity of the CarerQol in different groups of caregivers. These groups were (i) a heterogeneous group of caregivers that were member of informal care support centres in the Netherlands (chapter 2); (ii) caregivers of Dutch long-term care users (chapter 3); (iii) a heterogeneous sample of caregivers in the Netherlands from an online panel (chapter 4); (iv) parents of children with Autism Spectrum Disorders in the United States (chapter 5); and (v) caregivers of patients using palliative care in Australia (chapter 6). The psychometric properties feasibility, test-retest reliability and construct validity were investigated for both the CarerQol-VAS and the CarerQol-7D. The main findings will be discussed per psychometric criterion.

9.2.1 Feasibility of the CarerQol

For an instrument to be useful in practice, it is important that respondents are willing and able to answer the questions asked. In this thesis, feasibility was assessed by the proportion of complete responses. Almost all caregivers in chapters 2, 3, 5 and 6 answered all the questions on caregiving burden (CarerQol-7D) and happiness (CarerQol-VAS) of the CarerQol. Only between 4 and 8 per cent of respondents in these studies did not fill in all questions of the CarerQol. Previous work using written questionnaires also reported low levels of missing data on the CarerQol [110]. Explicit guidelines for acceptable levels of complete response, or other measures of feasibility, are in general lacking and therefore making firm inferences from the feasibility tests in this thesis is difficult. All in all, the results presented in this thesis appear to indicate a good feasibility of the CarerQol.

9.2.2 Test-retest reliability of the CarerQol

A second crucial criterion for measurement tools is that they produce the same results when administrated at different moments in time (all other things equal). This thesis addressed whether the responses on the CarerQol were stable over a two-week time interval among caregivers who reported no significant changes in their care situation. These test-retest reliability tests were performed among caregivers caring for patients using day care or those living in a long-term care facility in the Netherlands.

Main findings concerning test-retest reliability were twofold. First, judged by external standards of acceptable reliability [98, 155], the study reported in chapter 3 found very good stability of responses on the CarerQol-VAS measuring happiness over a two-week time period among caregivers of long-term care patients. Secondly, the stability of the seven burden dimensions of the CarerQol-7D was moderate to very good over the same time period. Overall, the test-retest correlations were of similar magnitude to those reported for other general life evaluation scales [302] and of those covering a specific domain of quality of life, like the EQ-5D or SF-6D health-related quality of life instruments [303-306]. Concluding, the CarerQol had reassuring levels of consistency of results over time and hence good test-retest reliability among caregivers active in long-term care.

9.2.3 Construct validation of the CarerQol

The last psychometric property studied in this thesis was construct validity. An important feature of measurement tools is that instruments indeed measure the construct they intend to measure. In the context of the CarerQol, it is crucial to investigate whether the CarerQol accurately reports the impact of caregiving on caregivers. Two methods have been used to study construct validity of the CarerQol: investigating correlations of the CarerQol with (i) other instruments measuring similar constructs (§9.2.3.1) and with (ii) contextual characteristics found to be important in the caregiving literature (§9.2.3.2). Conclusions on construct validation of the CarerQol will be presented in §9.2.3.3.

9.2.3.1 Correlation of CarerQol with similar instruments

The first approach to study construct validity used in this thesis was investigating the strength of correlation [98, 155] between the CarerQol and other instruments measuring constructs of subjective burden and well-being of caregivers. The CarerQol was compared to the (Expanded) Caregiver Strain Index (CSI; [105, 107, 110, 239-241]), the Self-Rated Burden scale (SRB; [11, 13, 34, 54, 105, 110]), the Carer Experience Scale (CES; [111, 112]), the Perseverance time (Pt; [169, 307-309]) instrument, the Family Quality of Life Scale (FQLS; [203]) and Process Utility (PU; [28, 33, 110]) in terms of convergent and discriminative validity, two types of construct validity. The following three hypotheses that were based on previous studies (e.g., [54, 105, 110]) were tested: (i) happiness of caregivers (CarerQol-VAS) is positively associated with positive caregiving experiences, (ii) positive caregiving dimensions of the CarerQol-7D are positively associated with positive caregiving experiences, and (iii) negative caregiving dimensions of the CarerQol-7D are negatively associated with positive caregiving experiences.

All five validation studies included in this thesis confirmed the first hypothesis that happiness of caregivers (CarerQol-VAS) was moderately to strongly positively associated with positive caregiving experiences measured on the CSI, SRB, CES, FQLS, and PU. Perseverance time was also associated with happiness of caregivers in the expected direction, but the association was relatively weak. The second hypothesis was that the positive caregiving dimensions of the CarerQol-7D (fulfilment from lending care and receiving support) would be positively associated with positive caregiving experiences on the other instruments (CSI, SRB, CES, Pt, FQLS, and PU). Most studies confirmed this hypothesis, but the results were less consistent, especially for the dimension support. Possibly, the two-directional effect of receiving support on caregivers' burden or well-being, as discussed in more detail in chapter 4, may explain these results. Finally, as posed in hypothesis iii, this thesis showed that in the large majority of caregiver samples, problems due to caregiving experienced by caregivers (CarerQol-7D) were negatively associated with positive caregiving experiences on the other instruments (CSI, SRB, CES, Pt, FQLS, and PU). The CarerQol-7D dimension "problems with daily activities" had the strongest association with these other instruments, CarerQol-7D dimensions "relational problems" and "financial problems" showed the weakest association.

Furthermore, convergent and discriminative validity tests investigated the association of the seven dimensions of caregiving (CarerQol-7D) with happiness of caregivers (CarerQol-VAS). In the range of caregiving samples included in this thesis, evidence was found that all seven problem dimensions of the CarerQol-7D had an impact on caregivers' well-being. Of these caregiving characteristics, mental health problems of caregivers seemed to be most important. Relational problems with the care receiver, financial problems, and the receipt of support with lending care showed the weakest associations with happiness of caregivers. Important to emphasize here is that certain caregiving problems were especially relevant in specific groups of caregivers. For example, difficulties with finances due to caregiving mainly seemed important for caregivers in low income groups.

9.2.3.2 Correlation of CarerQol-VAS with contextual characteristics of informal care

Construct validity of the CarerQol was also investigated by studying associations of the CarerQol with contextual factors which were previously shown to be important in explaining caregiver strain, such as the health status of care recipients, and included in the conceptual model of the impact of caregiving on caregivers used in this thesis (Figure 9.1). Here, this is referred to as clinical validity. Bivariate and multivariate analyses of happiness scores (CarerQol-VAS) were performed. The goal of bivariate analyses was to demonstrate differences in happiness scores among groups of caregivers known from the literature as a sign of construct validity of the CarerQol. In addition, multivariate analyses were used to explain differences in happiness scores among caregivers.

For bivariate analyses, groups of caregivers were distinguished based on the seven components of the conceptual model (Figure 9.1). Correlations of these seven components with happiness of caregivers (CarerQol-VAS) were found in the majority of bivariate analyses performed in this thesis. When studying the multivariate associations of the seven components of the conceptual model with happiness of caregivers (CarerQol-VAS) among the samples of caregivers in the Netherlands (chapters 2, 3 and 4), especially caregivers' health and the burden caregivers experience from caregiving were shown to be important. Chapter 4 also pointed to higher well-being scores of caregivers when relational ties were less tight between caregiver and care recipient, when caregivers had a paid work position, had higher income, were younger, and when they provided care to someone in better health. Many of these findings were replicated in the other samples of caregivers in the Netherlands (chapters 2 and 3). Important to note is that the influence of income of caregivers was only studied in chapter 4. Moreover, in chapters 2 and 3, in contrast to chapter 4, health of the care receiver was not significantly associated with the happiness of caregivers in the multivariate models, possibly due to heterogeneity in family ties in the caregiver samples or due to the relatively small sample sizes [178].

9.2.3.3 Conclusion construct validation CarerQol

Concluding, in general, the main hypotheses of this thesis were confirmed. As hypothesized, the CarerQol was associated with other instruments measuring similar constructs as the CarerQol. In addition, evidence was found in the majority of caregiver samples that the seven burden dimensions of the CarerQol-7D were significantly correlated with happiness of caregivers (CarerQol-VAS), in the expected direction. Concerning clinical validity, it can be concluded that in the majority of samples studied in this thesis the CarerQol-VAS captures differences in happiness scores among different groups of caregivers based on the conceptual model (see Figure 9.1). Although clear guidelines concerning the appraisal of findings regarding clinical validity are lacking, the results of this thesis are considered sufficient to label the clinical validity of the CarerQol as good.

Notwithstanding the fact that different concepts of the impact of caregiving were related to the CarerQol in the validation studies, in general the main hypotheses of this thesis hold. The results presented in this thesis thus suggest that the CarerQol validly measures the impact of caregiving on caregivers.

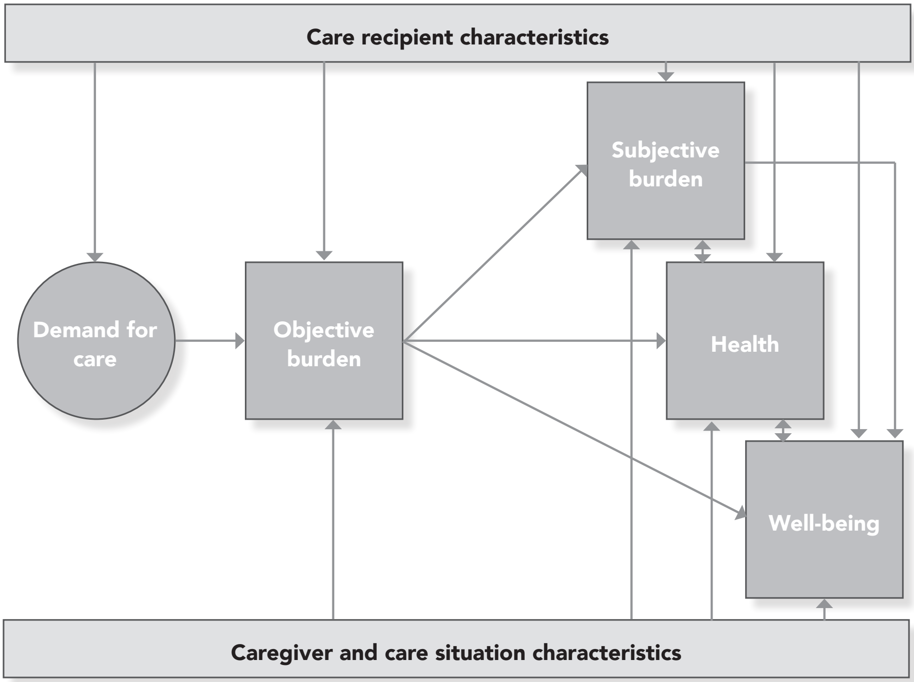


Figure 9.1 Conceptual model of the impact of caregiving on caregivers

9.3 Valuation of the CarerQol-7D states

To adequately inform policy makers about the overall impact of informal care on caregivers, this thesis reported utility based tariffs for the CarerQol-7D that can be applied in economic evaluations of health care interventions. This CarerQol-7D utility score is based on preferences from the general public for different caring states described with the CarerQol-7D and therefore is sensitive to differences in the severity of problems caregivers may experience. To obtain these CarerQol-7D utility scores, preference weights for the (different levels of the) two positive and five negative dimensions of caregiving of the CarerQol-7D were derived, using a discrete choice experiment (DCE) among the general public in the Netherlands (Chapter 7). In this DCE, respondents repeatedly had to choose between two hypothetical caregiving situations.

The results presented in chapter 7 show that people (as expected) preferred caregiving situations in which caregivers experienced fewer problems due to providing care and more fulfilment and support. Important dimensions of utility associated with an informal care situation were fulfilment from caring, physical health problems, relational problems and financial problems. A tariff providing standard utility scores for caregiving situations described by the CarerQol-7D was constructed using the relative preference weights of the CarerQol-7D dimensions.

Concluding, relative utility weights differ per dimension of caregiving. According to the general public in the Netherlands, fulfilment from caring, physical health problems, relational problems and financial problems had the most effect on informal care situations. Using this preference information, utility scores were computed for the different care situations that can be described using the CarerQol-7D. These utility scores correct for the importance of caregiving problems for caregivers' well-being and, hence, can be used in economic evaluations as a representation of care-related quality of life of caregivers informing policy makers on the impact of providing care on caregivers. Moreover, the CarerQol can be applied as an outcome measure serving as a weighted sum score of subjective burden of caregivers in more medically oriented research.

9.4 Overview of methods to include informal care in economic evaluations

This thesis furthermore reports how informal care can be included in economic evaluations of interventions in health care (chapter 8). As depicted in the conceptual model (Figure 9.1), caregiving can have a range of consequences. One of these consequences is that time needs to be invested by caregivers. In the absence of observed prices of informal care time, monetary valuation methods can be used to derive a value of informal caregivers' time investments. Resulting estimates of total costs can be included at the cost side of an economic evaluation. Moreover, informal care can affect caregivers' health or well-being and may result in subjective burden. These consequences of caregiving can be valued with non-monetary valuation methods in terms of health-related quality of life, well-being or care-related quality of life. Such valuations may be included at the effect side of an economic evaluation.

When choosing the most appropriate method to incorporate informal care in economic evaluations, three issues are especially important to consider: (i) the type of economic evaluation conducted (common cost-effectiveness or cost-utility analysis (CEA/CUA), cost-benefit analysis (CBA), or multi-criteria analysis (MCA)), (ii) the perspective of the economic evaluation (broad societal perspective or more restricted perspective, such as health care perspective), and (iii) the inclusion of informal care as a cost or as an effect. Economic evaluations conducted from a health care perspective can include health effects in caregivers as an effect of an intervention. The same valuation methodology as common for health effects in patients can be used. For CEA/CUA and MCA this would imply valuing health effects in terms of QALYs. In CBA, health effects of caregivers, like the effects in patients, should be expressed in monetary terms. The societal perspective allows the inclusion of broader consequences of caregiving in economic evaluations. In CEA/CUA adopting a societal perspective, QALYs of caregivers can be included at the effect side in combination with the valuation of time spent on caregiving at the cost side. To avoid double counting of health effects, the range of suitable valuation methods of caregivers' time should be restricted to those that merely value the time investment of caregiving, such as the opportunity cost method. Another strategy to include more consequences of caregiving than only health effects is the inclusion of the full impact of informal care in terms of costs. These methods can be included in economic evaluations adopting a societal perspective, regardless of the specific type of study (CEA/CUA/CBA/MCA). Several monetary valuation methods, aiming to value the full impact of informal care in monetary terms, exist, such as the willingness to pay and willingness to accept methods. Inclusion of the full impact of informal care at the cost side has important advantages in terms of the ease of practical application in research. However, when included as a part of the total costs of an intervention, policy makers are not explicitly informed about the consequences of caregiving. This is probably better achieved through inclusion of informal care at the effect side, such as discussed in terms of health-related quality of life. However, health effects represent only one of the many consequences caregiving might have. Other consequences, if measured in non-monetary terms, can only be included in a MCA. For instance, care-related quality of life of caregivers can be included in a MCA to pay explicit attention to the consequences of providing informal care in the context of economic evaluations of health care interventions. Important to emphasize is that the foregoing applies to economic evaluations of patient interventions. In the situation where interventions are evaluated that specifically target informal caregivers, care-related Qol can be used as the central outcome measure in CUA/CEA.

Concluding, several options are available for researchers to document the consequences of caregiving accurately, given the restrictions of the form and perspective of the economic evaluations conducted.

9.5 Strengths and limitations

This thesis studied psychometric criteria of the CarerQol (research question i), derived preferences for the CarerQol-7D states (research question ii), and provided an overview of methods to include informal care in economic evaluations (research question iii). This paragraph will address some strengths and limitations per research question.

An important strength of this thesis is that psychometric properties of the CarerQol were investigated in five studies using diverse groups of caregivers. These samples of caregivers differed on several aspects including (i) sample size, (ii) method of data collection, (iii) characteristics of caregivers and care recipients (e.g., their age and relationship between them), (iv) caregiving situation, (e.g., intensity of caregiving), and (v) country of study. In particular the CarerQol's feasibility and construct validation were repeatedly studied. Such replication of studies among samples in different contexts is important for obtaining evidence on psychometric properties of instruments. A difficulty in testing construct validity is that the CarerQol measures hypothetical constructs. The CarerQol-VAS measures happiness of caregivers and the CarerQol-7D describes the care situation and measures subjective burden. Although the lack of a gold standard for these hypothetical constructs makes it difficult to directly investigate validity of the CarerQol, a strength of this thesis is that the performance of the CarerQol was studied in relation to several commonly used and validated instruments. Furthermore, diverse instruments were also used to cover the seven components of caregiving of the conceptual model. An additional strength of this thesis is that different, but closely related, types of construct validity tests were used in this thesis, for instance convergent and discriminative validation. Although the use of a range of samples of caregivers, instruments and statistical tests is desirable for psychometric research, these factors also complicated the comparison of results found in the five validation studies of this thesis. The similarities in findings across the studies are encouraging.

Concerning limitations of this thesis, the specific way in which the psychometric properties of the CarerQol were studied deserves some attention. First, important to address is that some psychometric properties received no attention in this thesis, such as the responsiveness of the CarerQol. Another limitation is that the conclusion on the feasibility of the CarerQol is based on information from written questionnaires only, because no information on missing data on the CarerQol was available in the online panel of caregivers used in chapter 4. Moreover, information on reasons for non-response is not available. Secondly, test-retest reliability of the CarerQol was investigated in a modest sized sample of caregivers selected from a single long-term care facility. Despite the relatively small sample size, this sample had favourable characteristics for performing a test-retest reliability study. Most importantly, the care situation of caregivers was expected to be relatively stable over a short time frame, given the already severe and chronic nature of the health care needs of patients. Moreover, the intensity of caregiving was not very likely to change in these care situations, because most of the daily care was provided by professional caregivers from the long-term care facility. Consequently, responses on the CarerQol were less likely to change among this sample of caregivers in two weeks. Finally, construct validation of the CarerQol was investigated using a conceptual model of the impact of caregiving on caregivers. In this conceptual model different outcomes in caregivers and associated factors were depicted. Important to emphasize here is that some of the components of this conceptual model are likely to be interrelated, as indicated by the arrows in Figure 9.1. For example, the health status of caregivers is often one of the correlates of another caregiving outcome; subjective burden [11, 13, 22, 33]. This thesis avoids any causal claims, on for example whether subjective burden affects well-being or vice versa. Despite the fact that merely associations between different aspects of caregiving are considered, conclusions on factors associated with happiness of caregivers in this thesis may still be biased due to endogeneity problems. Furthermore, the results of this thesis may be biased, because the list of explanatory factors of caregivers' well-being used in this thesis is extensive, but not exhaustive [302, 310].

Some issues related to the second research question of the construction of the CarerQol-7D tariff deserve some further mentioning. In this thesis, the preferences of the general public for informal care situations were used to resemble the current practice of health utility elicitation studies. There is discussion in the literature on whose preferences to use in valuation studies; the values of the group affected (e.g., patients or –in this case- caregivers), professionals or the general public (e.g., [70]). An important advantage of using population values is the reduced influence of coping and adaptation. This thesis also showed some preliminary results of the application of the CarerQol-7D tariff. Among caregivers of palliative care patients in Australia, the CarerQol-7D utility score was associated with demand for care, duration of care, caregivers' health, and their age. The CarerQol-7D score was associated with fewer components from this thesis' conceptual model than CarerQol-VAS was (see paragraph 9.2.3.1). This can be expected, since the concept underlying the CarerQol-7D utility score, care-related quality of life, is less broad and more related to the process of providing informal care than general well-being of caregivers as measured with the CarerQol-VAS is.

Finally, this thesis provided an overview of methods to include informal care in economic evaluations. While this thesis showed that several methods are available to incorporate the impact of providing care on caregivers in cost-effectiveness research, this thesis did not study whether using these methods will actually affect cost-effectiveness recommendations. It is important to emphasize that this depends on several factors, such as the relevance of informal care in the specific care or disease context, the severity of the problems experienced by caregivers, the total costs of the intervention and the proximity of the incremental cost-effectiveness ratio to the threshold value the public or policy makers are willing to pay for the gains of the intervention [13, 85, 90, 264].

9.6 General conclusion

Different methods are available to incorporate informal care as a cost or as an effect in economic evaluations of interventions in health care. Including informal care at the effect side of an economic evaluation facilitates that the impact of caregiving will be explicitly noticeable for policy makers. This thesis showed that the CarerQol can be applied to measure and value the full impact of providing informal care on caregivers as an effect in economic evaluations. Specifically, the CarerQol can be used in multi-criteria analyses of patient interventions which are suitable for widening the range of consequences of interventions in health care. Moreover, the CarerQol can be used in CEA/CUA when interventions targeted at caregivers are evaluated. A recommendation to use the CarerQol in economic evaluations is facilitated by the favourable psychometric properties of the instrument. This thesis showed at least acceptable levels of the feasibility of the CarerQol among different groups of caregivers. Moreover, good test-retest reliability of the CarerQol among informal caregivers of long-term care users in the Netherlands was shown. Furthermore, the construct validity of the CarerQol was found to be favourable in multiple studies in different care settings and countries. Finally, this thesis facilitates the use of the CarerQol in economic evaluations by presenting preference-based tariffs to calculate utility scores for the CarerQol-7D.

9.7 Recommendations for research and policy

This thesis studied several psychometric properties of the CarerQol. Feasibility of the CarerQol was studied by analysing missing data on the CarerQol, but whether respondents are able and willing to answer the questions of the CarerQol could be studied further with other methods. For example, think-aloud studies can be applied to investigate whether respondents understand the questions, and whether these questions are appropriate to ask in specific groups of caregivers. For instance, it can be questioned whether it is meaningful to ask whether caregivers have relational problems with the care receiver when parents lend care to an ill child. Moreover, think-aloud studies can be used to investigate whether important caregiving problems are currently overlooked with the CarerQol instrument in specific groups of caregivers. For example, in long-term or palliative care patients often receive care from several health care professionals. It could be that an important part of caregiver strain in these groups consists of a lack of communication and coordination of care with professionals.

Although five studies investigated construct validation of the CarerQol, replication in other caregiving settings is desirable to study some specific findings of this thesis more in depth, such as the relatively weak association between the CarerQol-VAS and the perseverance time instrument. This finding could signal that these two instruments (not unexpectedly) measure different constructs.

Furthermore, more research on reliability, both including test-retest reliability and other reliability testing such as internal consistency, of the CarerQol is desired in different samples of caregivers to tests whether these groups of caregivers are similar enough to generalize findings. Moreover, further studies should also focus on other psychometric properties of the CarerQol, such as sensitivity to change.

As discussed before in paragraph 9.5, some components of the conceptual model are likely to be interrelated. Statistical adjustments for potential problems caused by endogeneity can be used to obtain more accurate information on the relationship between health, subjective burden and well-being in caregivers [310, 311]. Another solution is to obtain panel data on caregivers' well-being in future studies. Moreover, the conceptual model used in this thesis could be improved by including other factors known to be associated with for example caregiving strain. In social sciences it is usually not feasible to control for all (potential) influences. Nevertheless, factors not included in this thesis such as coping skills and personality traits are known to influence the way in which caregivers deal with problems due to caregiving [22]. In addition, when individuals evaluate their lives, psychological issues, such as aspirations in life and expectations about the future, seem to be important [302].

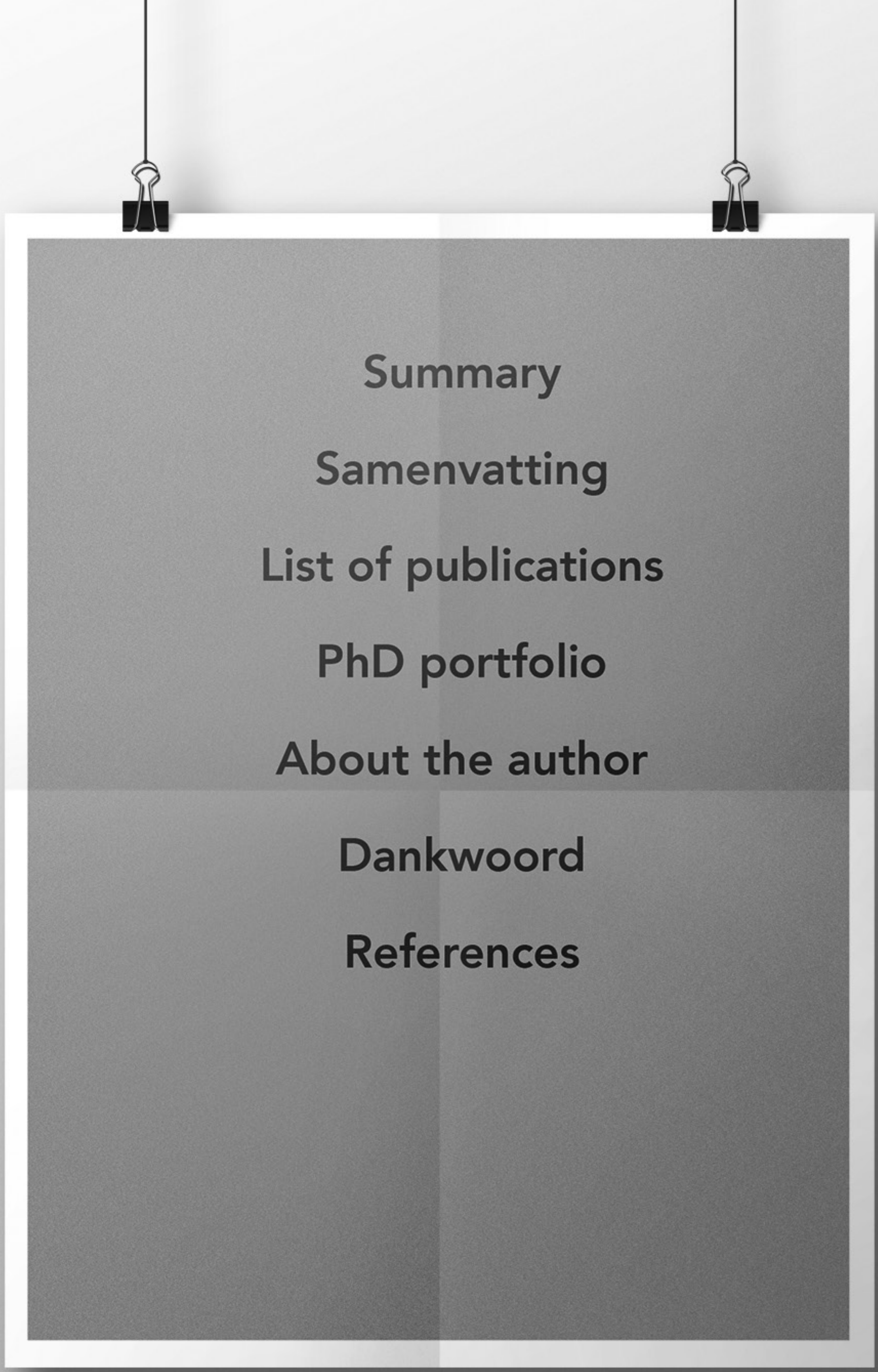
The CarerQol-7D tariff presented in this thesis was based on the preferences of the Dutch general public. Preferences for informal care situations may be influenced by cultural or institutional factors. Hence, it is recommended to perform valuation studies in other countries than the Netherlands. Moreover, the CarerQol-7D tariff could be calculated based on the preferences of caregivers. This provides the opportunity to study potential differences in utility related to informal care situations between the general public and caregivers.

This thesis focused on the impact of providing informal care on caregivers. An important, but often overlooked issue is how receiving informal care affects the care receivers. It could be hypothesized that the utility of care receivers is influenced by health or well-being effects in caregivers due to problems caregivers are faced with. More research into the interdependencies of patient and caregiver utility functions is needed to shed more light on this issue. Moreover, little is known on the preferences of (future) patients for receiving professional or informal care when confronted with care needs. More knowledge on how people wish to fulfil their care needs may help policy makers to arrange and plan long-term care arrangements in health care in line with these preferences.

Although this thesis focused on informal care in the context of patient interventions, it needs emphasis that certain caregiving problems which do not seem important for the majority of caregivers, can still be relevant in specific groups of caregivers. Further research could focus on subgroup analyses or apply statistical methods sensitive to the heterogeneity among caregivers to further demonstrate and explain this diverse impact of caregiving on caregivers. Moreover, general policy instruments may not be optimally effective in alleviating strain of caregivers if differences in subgroups exist. This implies that more tailored support interventions based on specific needs of caregivers may be required. For instance, financial support policies, such as caregiver allowances or tax deductibles, will probably be most effective in improving well-being of those caregivers who lack financial resources to cope with additional expenses of caregiving. When introducing such a measure more generally, public resources might be spent on caregivers for whom additional money not effectively improves well-being, as they do not experience financial strains. In the context of interventions for caregivers, policy makers and other professionals in the health care system should therefore be aware of and evaluate the problems and needs of specific caregivers. Consequently, they can offer (or advise on) the most appropriate type of support for those caregivers. At present, there are still many unanswered questions regarding the effectiveness of available caregiver support interventions [312-315]. Moreover, whether support programmes provide value for money is hardly ever evaluated [3]. Future research should therefore focus on which support interventions are most (cost)-effective and on why and in which care situations these interventions will benefit caregivers the most. This thesis underlines that methods are available that can serve as an outcome measure in such studies.

In relation to the main objective of this thesis, the most important recommendation is that researchers should include informal care in their economic evaluation. In this way, evidence on cost-effectiveness might guide policy makers to policy choices optimally benefiting society as a whole, including informal caregivers. The strain from and impact of providing informal care has been convincingly documented by now. Moreover, as this thesis shows, a range of methods and instruments is available, including the CarerQol, facilitating the inclusion of informal care in different types of economic evaluations. Most of these methods require little additional research effort, because instruments are available from the literature. Even if practical issues limit the additional collection of preference data on informal care costs or effects in economic evaluations, researchers can still include informal care in their cost-effectiveness calculations by applying monetary values of informal care already available in the literature. However, like in the case of the CarerQol, utility tariffs (or other preference weights) may not be available for all countries, so that tariff information from other countries needs to be used as an approximation.

Finally, policy makers and other commissioners of economic evaluation studies in health care should recognize the vital importance of informal caregivers in the care delivery to patients. Policy decisions in health care typically influence the lives of many individuals. This may be especially relevant in the context of long-term care where many family members and friends are involved in meeting the care demands of an ever increasing number of patients. Hence, policy makers should be aware that if they use cost-effectiveness information, this information should be adequate for public decision making. To increase the chance that policy makers are accurately informed of the consequences of their decisions on caregivers, they should make it compulsory that researchers include informal care in their evaluations, or provide valid reasons for excluding informal care. Guidelines for economic evaluations in health care should prescribe this and clearly indicate preferred methods for including informal care to increase consistent and comparable inclusion. Researchers should be encouraged to apply methods that include the full impact of informal care as much as possible. Moreover, caregivers should be adequately supported in their important task of providing care. This is also desirable given the serious problems in financing (long-term) care systems. In the end, acknowledging the importance of informal care in public policy making will not only benefit patients and their family and friends, but society as a whole. This thesis hopes to contribute to that aim.



Summary

Samenvatting

List of publications

PhD portfolio

About the author

Dankwoord

References

People confronted with illness, disability, or infirmity of old age, often rely on their family and friends for care. Providing such informal care may have a considerable impact on caregivers, both positive and negative. Policy makers are typically ill informed about these consequences of providing informal care. This thesis studied methods that facilitate the inclusion of informal care in economic evaluations of health care programmes. Economic evaluations that include informal care inform policy makers about the broader impact of interventions in society, and help them to allocate public budgets more efficiently, from a societal perspective.

In order to include informal care in economic evaluations, methods are needed. These methods should, first of all, describe the consequences of an intervention on caregivers in a feasible, reliable and valid way. Secondly, they should value the costs and benefits of informal care in a common unit, such as money or quality of life. This thesis addressed these two requirements for one specific instrument, the CarerQol. This instrument was designed to measure and value the full impact on caregivers of providing care for inclusion in economic evaluations. The CarerQol instrument is comprised of two parts. First, the CarerQol-7D records two positive (fulfilment and support) and five negative (relational problems, mental health problems, problems combining daily activities with care, financial problems and physical health problems) aspects of providing informal care. Secondly, the CarerQol-VAS measures well-being of caregivers in terms of happiness.

This thesis addressed three research questions. The first question concerned the feasibility, test-retest reliability and construct validity of the CarerQol instrument as a measure of the effect of caregiving on caregivers in different caregiving contexts. These psychometric properties were investigated in five studies using different samples of caregivers. Chapters 2, 3 and 4 used samples of caregivers from the Netherlands recruited through member registries of regional informal care support centres (chapter 2), a long-term care organization offering day and inpatient care (chapter 3) and a survey company (chapter 4). Chapters 5 and 6 used samples of caregivers from abroad: parents of children with an Autism Spectrum Disorder (ASD) in the United States (chapter 5) and caregivers providing care to patients who used palliative care services in Australia (chapter 6).

The feasibility of the CarerQol was studied in chapters 2, 3, 5 and 6. The extent to which respondents are willing and able to answer the questions asked was studied by investigating the proportion of complete responses to the CarerQol. The results showed that the large majority of respondents completed all the questions of the CarerQol. This led to the conclusion that the CarerQol had acceptable to good levels of feasibility among different groups of caregivers.

The reliability of the CarerQol was studied in chapter 3. Whether the CarerQol instrument reports the same results when it is administrated at different moments in time (all other things equal), was studied by comparing CarerQol scores collected with a two-week interval period. The results showed that scores were fairly consistent among caregivers without any significant changes in their caregiving situation. Given these results, test-retest reliability of the CarerQol among informal caregivers of long-term care users in the Netherlands was considered to be good.

The construct validity of the CarerQol was studied in chapters 2 to 6. The extent to which the CarerQol measures the construct it intends to measure was studied by investigating associations between the scores on the CarerQol instrument and scores on other validated instruments measuring outcomes in caregivers. In the majority of caregiver samples, the hypotheses about associations

between scores on the CarerQol instrument and scores on instruments of caregiver burden or well-being were confirmed. Moreover, expected differences were found in happiness scores (CarerQol-VAS) between subgroups of caregivers distinguished on the basis of characteristics of the caregiving situation found to be important in the literature. Overall, especially caregivers' health and the burden caregivers experience from caregiving were shown to be important in explaining happiness scores among caregivers. The results showed that the construct validity of the CarerQol in multiple studies among caregivers in different care settings and countries was favourable.
Reflecting on these results, the CarerQol appears to describe the consequences of caregiving in a feasible, reliable and valid way.

The second research question addressed the valuation of caregiving situations described by the CarerQol instrument. In chapter 7, a discrete choice experiment (DCE) was presented, designed to derive preferences of the general public in the Netherlands for caregiving situations defined by the three levels on the seven dimensions of the CarerQol-7D. The DCE showed that utility weights (also called tariffs) differed per dimension. In general, caregiving situations were preferred in which caregivers experienced fewer caregiving problems and more fulfilment from and support with caregiving. These tariffs for the CarerQol-7D can be used to value the impact of interventions on caregivers.

The third research question of this thesis addressed the range of methods available to include informal care in economic evaluations of interventions in health care. Chapter 8 showed that different methods can be used depending on the type and perspective of the economic evaluation, and that methods differed with respect to whether they measure and value a partial or the full impact on caregivers. When patient interventions are evaluated, the time investment of caregivers could be valued with common monetary valuation methods and the result could be included at the cost side of economic evaluations. Effects on caregivers' health could be measured using common health-related quality of life measures and included at the effect side of a cost-utility analysis, added to the health-related quality of life of patients. Well-being or care-related quality of life effects in caregivers could be measured using different instruments, for example the CarerQol, and could be included as one of the outcome measures in a multi-criteria analysis.
When caregiver interventions are evaluated, care-related quality of life of caregivers could serve as the main outcome in a cost-utility analysis.

Concluding, the research presented in this thesis showed that different methods are available to measure and value the impact of informal caregiving, and to include informal care in economic evaluations. If researchers, policy makers and commissioners of economic evaluation studies in health care wish to be informed about the broader impact of interventions in society, and to recognize the vital importance of informal care for the functioning of the health care system, they should strive to incorporate the impact of interventions on caregivers in their economic evaluations. Alternatively, guidelines for economic evaluations could prescribe this, so that decision makers in health care will receive accurate information about the societal costs and benefits of interventions, stimulating policy decisions benefiting society as a whole.

Mensen met een ziekte, beperking of ouderdomsklachten zijn vaak afhankelijk van de hulp van familie en vrienden. Het verlenen van deze mantelzorg kan een behoorlijk effect hebben op mantelzorgers, zowel positief als negatief. Beleidsmakers zijn vaak slecht geïnformeerd over deze gevolgen van het verlenen van mantelzorg. Dit proefschrift heeft methoden onderzocht die het mogelijk maken om mantelzorg op te nemen in economische evaluaties van interventies in de gezondheidszorg. Economische evaluaties die mantelzorg meenemen bieden beleidsmakers inzicht in de bredere maatschappelijke impact van interventies en ondersteunen hen om publieke middelen zo optimaal mogelijk te verdelen vanuit een maatschappelijk perspectief.

Het meenemen van mantelzorg in economische evaluaties vereist methoden die, ten eerste, de gevolgen van een interventie voor mantelzorgers op een gebruiksvriendelijke, betrouwbare en valide manier beschrijven. Ten tweede, deze methoden moeten de kosten en baten van mantelzorg waarderen in een gangbare eenheid, zoals geld of kwaliteit van leven. Dit proefschrift richtte zich op deze twee voorwaarden voor één specifiek instrument, de CarerQol. Dit instrument is ontwikkeld om de volledige impact van het verlenen van zorg op mantelzorgers te meten en te waarderen voor gebruik in economische evaluaties. Het CarerQol instrument bestaat uit twee delen. Het eerste deel, de CarerQol-7D, rapporteert twee positieve (voldoening en steun) en vijf negatieve (relationele, mentale, fysieke en financiële problemen en ten slotte problemen met het combineren van dagelijkse activiteiten met zorg) aspecten van het verlenen van mantelzorg. Ten tweede, de CarerQol-VAS meet welzijn van mantelzorgers in termen van geluk.

Dit proefschrift heeft drie onderzoeksvragen behandeld. De eerste vraag betrof de gebruiksvriendelijkheid, test-hertest betrouwbaarheid en construct (of begrips-)validiteit van het CarerQol instrument als meetinstrument van het effect van het verlenen van zorg op mantelzorgers in verschillende zorgsituaties. Deze psychometrische eigenschappen zijn onderzocht in vijf studies met verschillende groepen mantelzorgers. Hoofdstukken 2, 3 en 4 bestudeerden mantelzorgers uit Nederland die betrokken werden via regionale steunpunten voor mantelzorg (hoofdstuk 2), via een instelling voor langdurige zorg die dagopvang en intensieve zorg aan bewoners biedt (hoofdstuk 3) en via een onderzoeksbureau (hoofdstuk 4). Hoofdstukken 5 en 6 gebruikten steekproeven van mantelzorgers uit andere landen: ouders van kinderen met een stoornis in het autistisch spectrum uit de Verenigde Staten (hoofdstuk 5) en mantelzorgers van patiënten die gebruik maakten van palliatieve zorg in Australië (hoofdstuk 6).

De gebruiksvriendelijkheid van de CarerQol is onderzocht in de hoofdstukken 2, 3, 5 en 6. De mate waarin respondenten bereid zijn en in staat zijn om de CarerQol in te vullen is beoordeeld aan de hand van het percentage respondenten dat de CarerQol volledig invulden. De resultaten lieten zien dat een ruime meerderheid van de respondenten alle vragen van de CarerQol beantwoordden. Op basis van deze resultaten kon geconcludeerd worden dat de gebruiksvriendelijkheid van de CarerQol acceptabel tot goed was onder verschillende groepen mantelzorgers.

De betrouwbaarheid van de CarerQol is onderzocht in hoofdstuk 3. De vraag of de scores op het instrument hetzelfde zijn als het instrument op verschillende momenten wordt afgenomen (de overige omstandigheden gelijk blijvend) is onderzocht door het vergelijken van CarerQol scores die met een tussenperiode van twee weken verzameld zijn. De resultaten lieten zien dat de scores vrij consistent waren onder respondenten bij wie de zorgsituatie niet significant was veranderd. Deze resultaten leidden tot de conclusie dat de test-hertest betrouwbaarheid van de CarerQol goed was onder mantelzorgers van gebruikers van langdurige zorg in Nederland.

De construct validiteit van de CarerQol is bestudeerd in de hoofdstukken 2 tot en met 6. De mate waarin de CarerQol daadwerkelijk meet wat we verwachten dat het meet is onderzocht door het berekenen van de samenhang tussen de CarerQol scores en de scores op andere gevalideerde instrumenten die uitkomsten bij mantelzorgers meten. De hypothesen over deze samenhang tussen

de CarerQol scores en instrumenten die belasting of welzijn van mantelzorgers meten werden in de meerderheid van de hoofdstukken bevestigd. Verwachtte verschillen in de geluksscores (CarerQol-VAS) werden gevonden tussen subgroepen van mantelzorgers die verschilden met betrekking tot kenmerken van de mantelzorgsituatie die volgens de literatuur van belang zijn. In het algemeen beïnvloedden vooral de gezondheid van mantelzorgers en de belasting die zij ervaren hun welzijn. Deze resultaten lieten zien dat de construct validiteit van de CarerQol in verschillende populaties mantelzorgers in verschillende zorgsituaties en landen goed was.
Concluderend lieten de resultaten van de eerste onderzoeksvraag zien dat de CarerQol in staat lijkt om de gevolgen van mantelzorg op een gebruiksvriendelijke, betrouwbare en valide manier te beschrijven.

De tweede onderzoeksvraag richtte zich op de waardering van mantelzorgsituaties die beschreven worden met het CarerQol instrument. Hoofdstuk 7 rapporteerde de uitkomsten van een keuze-experiment dat de preferenties heeft achterhaald van het generale publiek in Nederland voor mantelzorgsituaties die beschreven zijn aan de hand van drie antwoordcategorieën op de zeven dimensies van de CarerQol-7D. De resultaten van dit experiment lieten zien dat de utiliteitsgewichten (ook tarieven genoemd) verschilden per dimensie. In het algemeen gaven respondenten de voorkeur aan mantelzorgsituaties waarin mantelzorgers minder problemen hadden, meer voldoening ontleenden aan het zorgen en meer steun hierbij ontvingen. Deze tarieven voor de CarerQol-7D kunnen gebruikt worden om de impact van interventies op mantelzorgers te waarderen.

De derde onderzoeksvraag van dit proefschrift behandelde de verschillende beschikbare methoden om mantelzorg op te nemen in economische evaluaties in de gezondheidszorg. Diverse methoden zijn beschikbaar voor de verschillende typen en perspectieven van evaluatie. Hoofdstuk 8 toonde aan dat de mate waarin deze methoden de volledige impact van het verlenen van zorg op mantelzorgers meten en waarderen verschilt. Het hoofdstuk wees er verder op dat de tijdsinvestering van mantelzorgers met gangbare monetaire waarderingsmethoden gewaardeerd kan worden en dan aan de 'kostenkant' van een economische evaluatie van interventies gericht op patiënten opgenomen kan worden. De gezondheidseffecten van mantelzorgers kunnen gewaardeerd worden met gangbare gezondheidsgerelateerde kwaliteit van leven instrumenten en aan de 'effect-kant' bij de gezondheidsgerelateerde kwaliteit van leven van patiënten opgeteld worden in een kosten-utiliteitsstudie. Uitkomsten bij mantelzorgers in termen van welzijn en zorggerelateerde kwaliteit van leven kunnen gemeten worden met andere instrumenten, zoals bijvoorbeeld de CarerQol, en meegenomen worden als één van de uitkomsten in een multi-criteria analyse. Ten slotte liet hoofdstuk 8 zien dat in kosten-utiliteitsstudies die interventies onder mantelzorgers evalueerden zorggerelateerde kwaliteit van leven van mantelzorgers als uitkomstmaat kan dienen.

Samenvattend, het onderzoek gepresenteerd in dit proefschrift heeft aangetoond dat er verschillende methoden beschikbaar zijn om de effecten van mantelzorg te meten en te waarderen, en om mantelzorg mee te nemen in economische evaluaties. Als onderzoekers, beleidsmakers en andere belanghebbenden van economische evaluaties in de gezondheidszorg een volledig beeld willen krijgen van het maatschappelijk effect van een interventie, en de waarde van mantelzorg voor het functioneren van het gezondheidszorgsysteem willen erkennen, dan zullen zij de beschikbare instrumenten moeten gebruiken om de impact van het verlenen van mantelzorg op te nemen in hun economische evaluaties. Richtlijnen voor economische evaluaties kunnen ook opnemen dat mantelzorg geïncludeerd moet worden, zodat beleidsmakers in de gezondheidszorg accurate informatie zullen ontvangen over de maatschappelijke kosten en baten van een interventie. Dit zal beleidsmakers stimuleren om beleidsbeslissingen te nemen die bijdragen aan het welzijn van de samenleving als geheel.

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PhD portfolio Renske Hoefman

PhD training

2013	Best Worst-Scaling, The Centre for the Study of Choice, Sydney
2011	Introduction to Rasch Analysis, Psychometric Laboratory for Health Sciences, University of Leeds
2011	Modeling Health Care Costs and Counts Workshop, International Health Economics Association, Toronto
2011	Executive Course Discrete Choice Experiment, The University of Sydney, Rotterdam
2011	Basisdidactiek, Risbo, Research-Training-Consultancy, Erasmus University Rotterdam
2010	Writing Academic English for iBMG, Academic Language Centre, Leiden University
2010	Klaar in vier jaar, Brigitte Hertz, Rotterdam
2010	Discrete Choice Modeling, Centre for Microdata Methods and Practice, Institute for Fiscal Studies, London
2010	Advanced Course in Applied Health Economics, Methods for the analysis of categorical dependent variables, Centre for Health Economics, The University of York
2008	Erasmus Summer Programme, Regression Analysis, NIHES Research Training in Medicine and the Health Sciences, Rotterdam
2008	Tutortraining Probleemgestuurd Onderwijs, institute for Psychology, Erasmus University Rotterdam

Teaching

2007- 2010	Socio-medical sciences, practicum, bachelor program Health Sciences, institute of Health Policy and Management, Erasmus University Rotterdam
2008 – 2013	Introduction course in health care, practicum, bachelor program Health Sciences
2008 – 2013	Supervisor and co-evaluator for bachelor and master theses, institute of Health Policy and Management, Erasmus University Rotterdam
2009	Introduction week students, practicum, bachelor program Health Sciences
2011-2013	Patient preferences in the delivery of health care, practicum and lecture, Master program Health Economics, Policy and Law, Erasmus University Rotterdam
2012-2013	Health Technology Assessment, lecture, Master program Health Economics, Policy and Law
2012-2013	Writing and research skills for premaster students, practicum, premaster program institute of Health Policy and Management, Erasmus University Rotterdam
2013	EuroQol Discrete Choice Course, practicum, Amsterdam

Conferences

Podium presentations

2014	Dutch Conference on Public Health, Rotterdam
2013	9th World Congress on Health Economics, International Health Economics Association, Sydney
2012	19th Annual Conference of the International Society for Quality of Life Research, Budapest

2012	9th European Conference on Health Economics, Zürich
2011	8th World Congress on Health Economics, International Health Economics Association, Toronto
2011	3rd Lowlands Health Economic Study Group, Soesterberg
2010	8th European Conference on Health Economics, Helsinki
2009	1st Lowlands Health Economic Study Group, Berg en Terblijt
2009	2nd Joint European Public Health Conference, Lódz
2008	1st Joint European Public Health Conference, Lisbon

Poster presentations

2010	8th European Conference on Health Economics, Helsinki
2009	Annual European International Society for Pharmacoeconomics and Outcomes Research (ISPOR) Congress, Paris

Presentations at other meetings

2011	Seminar University of Sheffield/ Erasmus University Rotterdam
2013	InterTASC workshop, organised by KSR, Erasmus University Rotterdam and Maastricht University
2013	'Coffee & Research', VU University Medical Center
2012	Outcomes beyond the QALY, joined NVTAG/ZonMw/iBMG symposium
2011	Research meeting Informal care, Movisie
2011-2010	Seminar institute of Health Policy and Management

Other meetings and workshops

2013	International Choice Modeling Conference, Sydney
2013	Expert meeting 'Informal Care', organised by the Netherlands Institute for Social Research and the ministry of Health, Welfare and Sport, The Hague
2012-2010	Lowlands Health Economic Study Groups
2010	5th National Informal Care Meeting, Utrecht
2010	Health Rationing Conference, Erasmus Observatory on Health Law, Rotterdam

International collaboration and visits

US	Prof. J. Mick Tilford, University of Arkansas for Medical Sciences
	Dr. Nalin Payakachat, University of Arkansas for Medical Sciences
	Dr. Karen Kuhlthau, Massachusetts General Hospital
UK	Dr. Hareth Al-Janabi, University of Birmingham
Australia	Prof. John Rose, University of Sydney
	Prof. Julie Ratcliffe, Flinders University
	Prof. David Currow, Flinders University
	Dr. Nikki McCaffrey, Flinders University

Renske Hoefman (1983) studied Health Sciences at the Erasmus University in Rotterdam (2002-2005). In 2007, she obtained a masters degree in Health Economics, Policy and Law from the Erasmus University in Rotterdam. In 2007 and 2008, Renske worked as a researcher on informal and elderly care at the Netherlands Institute for Social Research (SCP). She continued her work on caregivers at the institute of Health Policy and Management at the Erasmus University in Rotterdam in 2008 and specialized in the role of informal care in economic evaluations and questions on validity and reliability of measurement tools.

Besides her academic work, Renske organizes activities for children and young adults in her community and she is a member of the editorial staff of the organisation for patients with Crohn’s disease and ulcerative colitis in the Netherlands.

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